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Spring-mediated sagittal craniosynostosis treatment at the Children's Hospital of Philadelphia: technical notes and literature review

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OBJECT Sagittal craniosynostosis has been treated using both cranial remodeling techniques and modification of the sagittal strip craniectomy. A more recent technique is to implant springs in conjunction with a suturectomy to transversely expand the parietal bones to accommodate the growing brain. In this paper the authors describe and evaluate several modifications to the spring-mediated cranioplasty (SMC) technique, most notably use of an ultrasonic scalpel to limit dural dissection and maximize opening of the stenosed suture by placement of multiple spring devices. In addition, the literature is reviewed comparing SMC to other surgical treatments of sagittal synostosis.

METHODS The authors retrospectively reviewed patients who presented to the Children's Hospital of Philadelphia with a diagnosis of sagittal synostosis from August 2011 to November 2014. A pooled data set was created to compare our institutional data to previously published work. A comprehensive literature review was performed of all previous studies describing the SMC technique, as well as other techniques for sagittal synostosis correction.

RESULTS Twenty-two patients underwent SMC at our institution during the study period. Patients were 4.2 months of age on average, had a mean blood loss of 56.3 ml, and average intensive care unit and total hospital stays of 29.5 hours and 2.2 days, respectively. The cranial index was corrected to an average of 73.7 (SD 5.2) for patients who received long-term radiological follow-up. When comparing the authors' institutional data to pooled SMC data, blood loss and length of stay were both significantly less ($p = 0.005$ and $p < 0.001$, respectively), but the preoperative cranial index was significantly larger ($p = 0.01$). A review of the SMC technique compared with other techniques to actively expand the skull of patients with sagittal synostosis demonstrated that SMC can be performed at a significantly earlier age compared with cranial vault reconstruction (CVR).

CONCLUSIONS The authors found that their institutional modifications of the SMC technique were safe and effective in correcting the cranial index. In addition, this technique can be performed at a younger age than CVRs. SMC, therefore, has the potential to maximize the cognitive benefits of early intervention, with lower morbidity than the traditional CVR.

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KEY WORDS sagittal synostosis; spring-mediated cranioplasty; craniosynostosis; cranial vault remodeling

Sagittal synostosis is the most prevalent type of craniosynostosis, with an incidence of 1 in 4200 births.[10] Premature closure of the sagittal suture is associated with the poor aesthetics of an elongated head shape with frontal and occipital bossing, and has been associated with increased intracranial pressure,[21] disruption in blood flow,[6] and poor neurological development. Early treatment of synostosis allows for correction of cosmetic deformities and may have a positive impact on neurocognitive development.

The earliest-described surgical treatment of sagittal craniosynostosis involved removal of the fused suture and was performed by Lannelongue,[14] followed by Lane[13] in the 1890s. These early approaches, focused on removing the suture, had a high level of failure[19] and soon gave way to more extensive cranial vault remodeling (CVR) tech-

ABBREVIATIONS CHOP = Children's Hospital of Philadelphia; CVR = cranial vault remodeling; ICU = intensive care unit; SMC = spring-mediated cranioplasty.

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niques. Current remodeling procedures range from sagittal synostectomy[26] or sagittal synostectomy with parietal craniectomies,[1] to more complex CVR in 1 or 2 stages.[7,11] There is an increasing body of evidence to suggest that from a neurocognitive standpoint, outcomes are improved when intervention occurs early (less than 6 months of age) and with either CVR or active expansion after suturectomy.[9] However, a large procedure at a younger age poses considerable risks, including prolonged operative time, increased blood loss, and longer hospital stays.

In an attempt to avoid some of the morbidity of CVR, modern minimally invasive techniques have been developed that release the synostosed suture, increase intracranial volume, and improve cranial morphology. One of the earliest techniques involved endoscopic strip craniectomy followed by extensive helmet orthosis use to achieve similar results to CVR.[12] Another technique, first performed by Lauritzen et al., uses a minimal strip craniectomy followed by internal distraction with springs to achieve adequate cosmetic results.[15]

Since its introduction, spring-mediated treatment of sagittal synostosis has been evaluated at multiple institutions with reasonable results.[3,4,8,16,17,22,25,27] At the Children's Hospital of Philadelphia (CHOP), we use a modification of the spring-mediated cranioplasty (SMC) technique to treat sagittal synostosis without the prolonged use of an external orthosis. In this report we present our recent results of SMC, including our modifications of the technique. In addition, we review the literature concerning the rationale for the spring-mediated sagittal synostosis procedure.

Methods

Data Collection

Following institutional review board approval, we retrospectively reviewed the records of all patients who had undergone spring-mediated surgery for sagittal synostosis at the CHOP during the period from August 2011 to November 2014. These patients were compiled from a craniosynostosis database approved by the institutional review board. Factors including age, pre- and postoperative cranial index values, length of stay in the intensive care unit (ICU), length of stay in the hospital, and estimated blood loss were reported and compared with the corresponding values from the pooled data. For this data set, only the papers providing total number, mean, and standard deviation of the relevant factors were included for statistical analysis.[8,16,22,25,27] The cranial index was collected preoperatively using CT scans to report the ratio of maximal biparietal distance over maximal occipitofrontal distance multiplied by 100. Postoperatively, cephalograms were used to obtain the biparietal distance and occipitofrontal distance of patients who had springs implanted for more than 2 months. A 2-sample t-test was used to compare the values obtained from the literature to those from CHOP's clinical data. Representative patient photos are presented in Fig. 1B and C.

Operative Technique

Preoperative Helmeting

In our early experience, we noted that the dolichocephalic deformity worsened considerably in patients between the time of their initial evaluation (often at 4–6 weeks of age) and surgical treatment (age 3–4 months). Sood and colleagues reported the successful normalization of dolichocephaly in 4 patients treated with cranial molding helmets prior to cranial vault reconstruction at 6 months of age.[20] Therefore, we normally advise initiating helmet orthosis treatment shortly after the initial consultation and continuing until the time of the surgical procedure. Since introducing this modification we have noted less deterioration in head shape in patients at the time of surgery. Furthermore, we have noted an improvement in head shape, particularly in the more phenotypically severe patients, i.e., those with significant saddling, severe occipital bossing, severe frontal bossing, and cranial indices less than 67.

Surgical Treatment

In the operating room, the patient is turned prone onto a cerebellar head holder. In some instances, the patient may be placed supine if a portion of the metopic suture also requires removal. Two linear incisions are planned just posterior to the anterior fontanelle and just anterior to the posterior fontanelle (Fig. 1A). The skin is normally infiltrated with a solution of bupivacaine hydrochloride and epinephrine. In most instances the skin is incised with a PEAK PlasmaBlade (Medtronic) to reduce scarring and bleeding. Next, a subcutaneous tunnel is made between the 2 incisions anteriorly and posteriorly. The anterior fontanelle is undermined and a bur hole is placed at the site of the posterior fontanelle. This bone opening is widened to just beyond the width of the craniectomy. The dura is then stripped from the bone, and an approximately 1.5-cm strip of bone is removed along the site of the fused sagittal suture using a Sonopet ultrasonic knife with a Nakagawa serrated knife tip (Stryker). This craniectomy can also be performed using a B1 bit on a drill with a footplate or a large pair of scissors. If desired, the dural stripping can be performed with the assistance of the endoscope. However, we have found that with the use of the Sonopet, the endoscope is normally not needed.

Following sagittal suturectomy, 2 to 3 springs are placed ranging from 6 to 11 N in force. The posterior spring is placed 1 cm anterior to the posterior fontanelle, and the anterior spring is placed 2 cm posterior to the anterior fontanelle. Care is taken in the selection of the force of the spring with respect to the age of the patient, thickness of the bone, degree of synostosis present, and amount of correction needed. A preoperative CT scan can be useful for this selection process (Fig. 2A). The goal is to maximize force without tearing through bone, and we have found springs in the 6–11 N range to be both safe and effective. If the intraoperative effect on the middle vault after placement of 2 springs was deemed inadequate by the senior craniofacial surgeon, a third spring was added in the midpoint of the suturectomy. The patient's strip craniectomy bone is morselized and then placed back into the sagittal strip area as cranial bone graft. The heads of the baseplates of the springs are sutured to the periosteum surrounding the strip craniectomy with interrupted 5-0 chromic sutures to decrease baseplate mobility. Hemostasis can be assisted with the use of some hemostatic matrix.

Postoperatively, cranial radiographs are often obtained

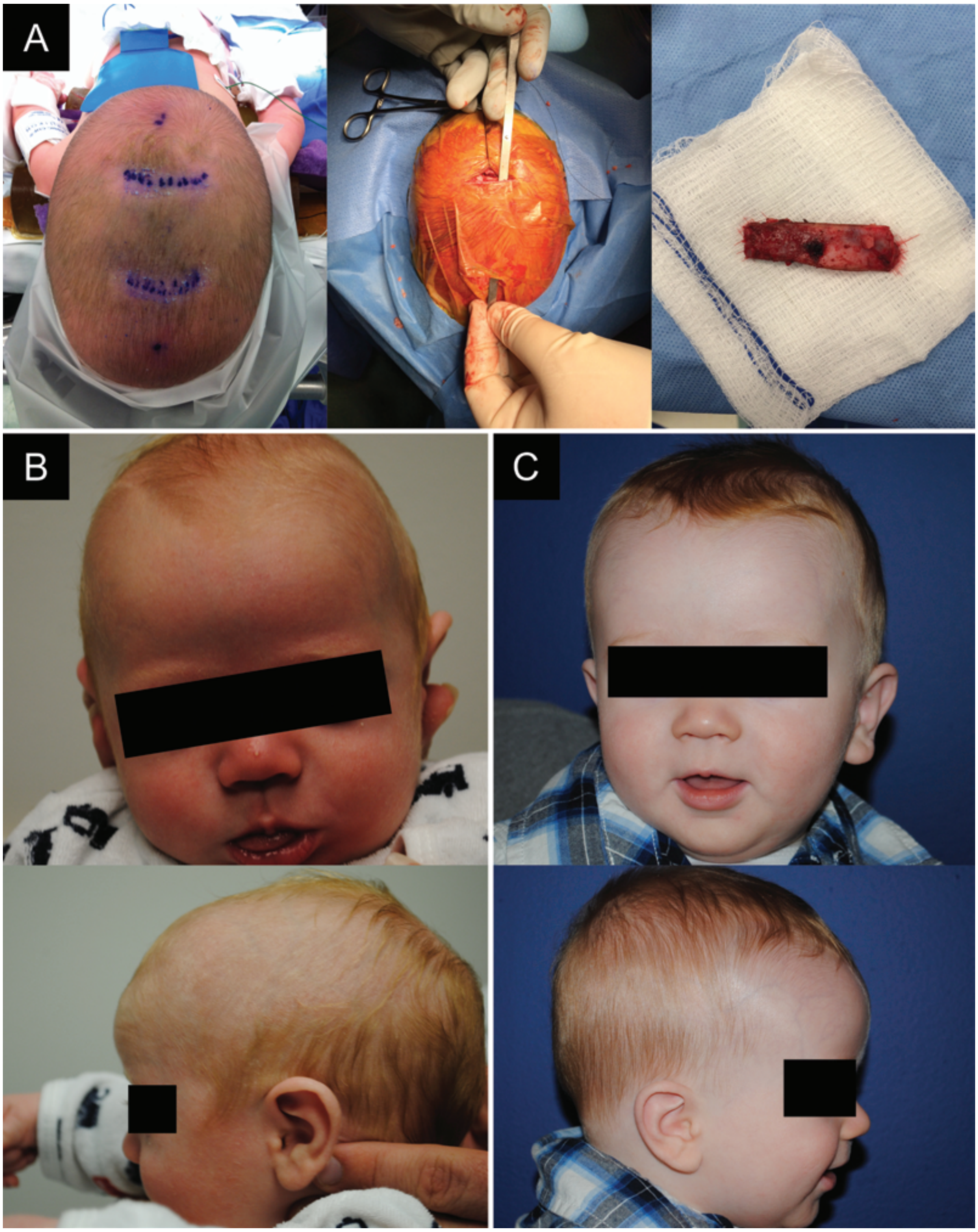

FIG. 1. Representative SMC photos of a single representative patient. **A:** Intraoperative images showing the patient supine with single dots representing the anterior and posterior fontanelle and dotted lines representing the planned incision *(left)*; Gigli saw guide placed into the epidural dissection plane *(center)*; and bone from the sagittal craniectomy *(right)*. **B:** Preoperative photos of the same patient showing anterior *(upper)* and lateral *(lower)* views. **C:** Postoperative photos at 6 months of the same anterior *(upper)* and lateral *(lower)* views.

to establish the location of the springs (Fig. 2B). Selected patients are treated with helmet orthosis for short periods of time after the surgical procedure. The patients then return to the operating room in 16–20 weeks for removal of the device as an outpatient procedure.

Literature Review

We performed a PubMed key word Boolean search using "[craniosynostosis OR sagittal synostosis]" AND "surgery" AND "spring", which yielded 51 articles. Of those articles, 7 studies[3,4,8,17,22,25,27] were found that reported at least 5 cases of sagittal synostosis that were managed by SMC. The references of each paper were also reviewed to search for other relevant studies. These 7 studies were included in our systematic review of spring-mediated surgical management of sagittal synostosis. The data collected from these studies was pooled to study patient age, surgical outcomes (measured by the cephalic index), duration of surgery, length of hospital/ICU stay, and other comorbidities (Table 1). The studies that specifically compared

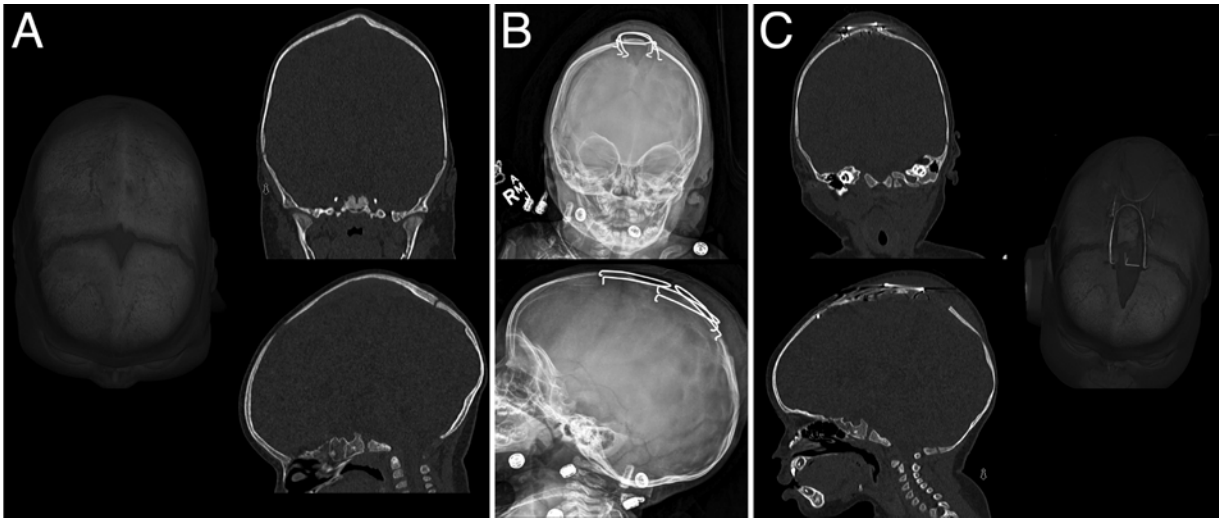

FIG. 2. Pre- and postoperative imaging of the same patient as in Fig. 1. **A:** Preoperative 3D CT coronal *(upper right)* and sagittal sections *(lower right)* and 3D-CT reconstruction *(left)*. **B:** Coronal *(upper)* and sagittal *(lower)* views of the immediate postoperative cephalogram. **C:** Postoperative 3D-CT coronal *(upper left)* and sagittal sections *(lower left)*, and 3D reconstruction showing spring positions *(right)*.

spring-mediated surgeries to other procedures for treating sagittal craniosynostosis[3,4,8,17,22,25,27] were reported separately in Table 2. All reported means were calculated as weighted averages from the pooled data.

Results

CHOP SMC Compared With Pooled Literature SMC

Patients from the CHOP had a mean age of 4.2 months (SD 1.8 months), which was not statistically different from the pooled data set[8,22,25,27] ($p = 0.86$; Table 1). In the CHOP cohort, 54.5% of the patients received 3 cranial springs and the remainder received 2 springs. The average spring strength was 9.0 N (SD 1.36 N) for the anterior spring, 9.25 N (SD 1.45 N) for the midsagittal spring, and 10.05 N (SD 1.22 N) for the posterior spring. The mean blood loss was significantly less in the CHOP cohort compared with the pooled data, at 56.3 ml (SD 35.3 ml, $p = 0.005$). The CHOP patients spent an average of 2.2 days in the hospital, which was significantly less than the 5.2 days of the pooled data ($p < 0.001$). There was a significantly longer stay in the ICU at the CHOP ($p = 0.03$). There was not a significant difference in operative time ($p = 0.83$). It should be noted that it has been the general practice to place all patients with craniosynostosis in the ICU postoperatively as was customary prior to the introduction of SMC, when patients were treated with CVR. It is the belief of the senior authors that this level of care is likely not needed for the majority of the patients treated with SMC. The preoperative cranial index index was significantly higher for CHOP patients relative to the pooled data set ($p = 0.01$), but there was no significant difference in postoperative cranial index with a mean of 73.7 (SD 5.2, $p = 0.92$; Table 1). The springs were removed an average of 128.4 days later (SD 29.7 days). On average, the spring removal procedure took 44.9 minutes (SD 17.6 minutes), yielded an estimated blood loss of 20 ml (SD 28.4 ml), and resulted in a mean postoperative hospital stay of 0.9 days (SD 0.3 days).

Review of the Literature

The average age of all patients treated with SMC in the pooled data of studies from the literature was 5 months (Table 2). The mean blood loss was 71.1 ml and the average length of surgery was 61.8 minutes. The average amount of time spent in the ICU was 11.8 hours, while the average postoperative stay was 3.07 days. Patients overall showed a 10.4% increase in the cephalic index from 67.5 preoperatively to 74.5 postoperatively. Springs were maintained in the patients for an average of 5.1 months. The average reported follow-up time was 27.5 months, at which point the average cranial index increased to 75.2 (Table 2).

We also reviewed comparisons of surgical techniques in the literature. The surgical procedures, relevant citations, average age of patients, blood loss, surgery time, postoperative stay, ICU stay, preoperative cranial index, and postoperative cranial index are shown in Table 3.

SMC Versus Pi-Plasty

Two studies[8,27] compared the outcome of SMC with the pi-plasty procedure. In both papers, the authors reported a younger mean age ($p < 0.001$, Windh et al.;[27] $p = 0.002$, Guimarães-Ferreira et al.[8]) as well as a significant reduction in blood loss ($p < 0.001$ and $p = 0.003$, respectively) and postoperative stay ($p < 0.001$ and $p < 0.013$, respectively) for SMC patients relative to pi-plasty patients. Windh et al.[27] found a significant decrease in duration of surgery ($p = 0.0103$) as well as in ICU stay ($p = 0.03$) for SMC patients. Pre- and postoperative cranial index values showed no significant differences in the study of Guimarães-Ferreira et al.[8] However, Windh et al.[27] reported a significant difference in postoperative cranial index, with pi-plasty resulting in a higher cranial index ($p = 0.0128$).

TABLE 1. A comparison of clinical data from the CHOP with data in the literature

Cohort	Age (mos)	Blood Loss (ml)	Spring Removal Blood Loss (ml)	Surgery Time (min)	Spring Removal Surgery Time (min)	Postop Stay (days)	Spring Removal Postop Stay (days)	ICU Stay (hrs)	Preop CI	Postop CI (>2 mos)
Pooled*										
Mean	3.5	84.9		91.9		5.2		23.2	66.4	73.9
SD	0.6	41.7		32.7		2.1		5.7	4.1	3.6
No. of patients	37	68		37		37		37	78	78
CHOP										
Mean	4.2	56.3	20	93.7	44.9	2.2	0.9	29.5	69.9	73.7
SD	1.8	35.3	28.4	23.8	17.6	1.4	0.3	14.3	5.3	5.2
No. of patients	22	20	13	16	13	20	20	22	19	7
p value	0.86	0.005		0.83		<0.001		0.03	0.01	0.92

CI = cranial index.
* Pooled data from the following studies: 8, 22, 25, and 27.

SMC Versus CVR/Cranial Expansion and Strip Craniectomy With or Without Parietal Barrel Staving

David et al.[3] directly compared the outcomes of their SMC patients with those treated with CVR. Their SMC patients were significantly younger ($p < 0.001$), experienced less blood loss ($p < 0.001$), had shorter operations ($p < 0.001$), and had shorter postoperative hospital ($p < 0.001$) and ICU stays ($p < 0.001$) than CVR patients. Patients treated with CVR had significantly lower preoperative cranial index values ($p < 0.036$), while postoperative cranial index values did not show a statistically significant difference. Mackenzie et al.[17] used an ANOVA to compare SMC outcomes with those of strip craniectomy, strip craniectomy with parietal barrel staving, and strip craniectomy with bioabsorbable cross-struts. The authors examined age, blood loss, length of operation, and hospital stay. They found a significant difference in duration of surgery ($p < 0.01$) between SMC and the other procedures, with SMC demonstrating shorter operative times. The other factors did not reach statistical significance. Two studies[17,22] compared SMC to strip craniectomy with parietal barrel staving. Taylor and Maugans[22] reported significantly less blood loss ($p < 0.001$), surgery time ($p = 0.002$), and postoperative stay ($p = 0.009$) for SMC patients relative to patients undergoing strip craniectomy with parietal barrel staving. They saw no significant difference in ICU stay, preoperative cranial index, or postoperative cranial index between these procedures.

Discussion

From the current data set, we have shown that the CHOP modification of the SMC technique can be safely and effectively performed in children less than 6 months of age. Postoperative course was maintained at 1 postoperative day in the ICU followed by 1 additional day in the hospital. Blood loss was also minimal due to technique changes. In addition, the cranial index was corrected to normal for most of the SMC patients at CHOP.

Based on our review of the literature, the SMC practice at CHOP has yielded similar cranial index correction when compared with other SMC patient reports.[3,4,8,16,17,22,25,27] Changes in techniques compared with those previously described by Lauritzen et al.[16] and his trainees may have led to the significant decrease in estimated blood loss and in length of postoperative stay noted in our analysis. The use of 2 small incisions over the fontanelle instead of a larger S-shaped incision may decrease overall blood loss and subsequent morbidity. Additional technique changes performed at CHOP may not be evident in the objective values measured, but nonetheless make the SMC a safe and effective surgery. One such change in technique is the use of the Stryker Sonopet single-sided Nakagawa serrated knife tip to perform the craniectomy. Previous reports have used the Misonix BoneScalpel (a double-edged ultrasonic knife) or the Depuy Synthes piezoelectric system (a short large-tooth blade) with incidences of dural tears in a small cohort of patients.[2] In our experience, the Sonopet knife has provided the same advantages of the previously reported ultrasonic knives, including endoscopic use and decreased blood loss, but with a better safety profile.

Another difference between our analysis and reports in the literature is the relatively high preoperative cranial index observed in our patients. This high value can be explained by the use of helmets prior to SMC in some of these patients. These helmets were in place from the time of initial examination until surgery. With helmets, improvement and prevention of dolichocephalic decline has been observed specifically in those with significant saddling, severe occipital bossing, severe frontal bossing, and cranial indices less than 67. The above-average preoperative cranial index noted in this study reflects the practice of helmet placement. Although the cephalic index is improved with preoperative orthosis use, at the age of 3–4 months the cranium still has yet to grow approximately 25% in circumference and 50% in volume to maturity.[18] In the setting of sagittal suture fusion, further biparietal expansion from this point would likely be impeded; for this reason, suturectomy is a standard component of all current treatments to release the constricting suture. We view preoperative orthotic use as a temporizing measure to limit worsening of the deformity prior to suture release and cranial vault expansion; we do not believe that it alone would facilitate an optimal cranial volume and contour at

TABLE 2. Pooled data set of studies reporting SMC

Authors & Year	No. of Patients	Age (mos)	Blood Loss (ml)	Surgery Time (min)	Postop Stay (days)	Duration of Springs (mos)	ICU Stay (hrs)	Preop CI	Postop CI	% CI Change	Follow-Up (mos)	Follow-Up CI
Taylor & Maugans, 2011	7	3.7	24.3	46	1.4		16.8	64.9	78.9	21.6		
Windh et al., 2008	20	3.5	170	104	6	6	25	66.57	70.57	6.0		
Guimarães-Ferreira et al., 2003	10	3.2		100	6.4		23.9	65.15	71.23	9.3		
Lauritzen et al., 2008	35	3.8	143	97	5	7	25	67	74	10.4	6	74
van Veelen & Mathijssen, 2012	41	5.8	53.8					66.8	75.4	12.9	12	73.7
David et al., 2010	75	5.7	25	30	0.93	4	0	69	75.4	9.3	46	76.6
Mackenzie et al., 2009	9	7		66	4.4							
Mean*		5.02	71.1	61.8	3.07	5.12	11.78	67.53	74.53	10.4	27.5	75.21
No. of patients	197	197	178	156	156	130	147	188	188	188	151	151

* Means calculated as weighted averages from the pooled data.

skeletal maturity. To date there is limited data on treating patients with a helmet only, and standard treatment is surgical intervention.[20]

Patients whose intraoperative midvault expansion after placement of 2 springs was inadequate, as judged by the senior craniofacial surgeon, underwent the placement of a third spring. While it may be possible to associate improved comorbidity outcomes with the relatively low amount of required correction in the CHOP patients, it is important to clarify that, unlike in more drastic surgeries such as CVR, recovery time is not directly dependent on the degree of correction needed for the SMC procedure. Regardless of the starting level of deformity, the SMC requires essentially the same level of intervention in the skull (strip craniectomy with spring placement), and therefore our study of SMC patients at CHOP suggests that the improved outcomes may be, in part, attributable to the precautionary techniques used in addition to the methods practiced at other institutions.

When compared directly to other minimally invasive or total cranial remodeling techniques in the literature, SMCs have had similar outcomes of decreased operative morbidity. In the current literature, SMC has been directly compared with the pi-plasty procedure,[8,27] sagittal strip craniectomy with and without parietal barrel staving,[17,22] strip craniectomy with bioabsorbable cross-struts,[17] and CVR.[3,4]

When compared with the pi-plasty procedure, SMC appears to have comparable results as far as the cranial index. In addition, SMC patients had significantly shorter hospital stays and significantly fewer blood transfusions. These results were confirmed by 2 separate studies from the same surgical center.[8,27] Although statistical significance was not reached in 1 study, SMC was also associated with shorter operative time and a shorter ICU stay for patients. Additionally, both papers included subjective parental questionnaires about cosmetic results, and the parents of patients who underwent SMC were satisfied with the outcomes. Windh et al. attributes the differences in postoperative cranial index to the mechanics of the pi-plasty procedure, where the shortened cranial length actively puts a limitation on cranial height. Both techniques, however, still achieved cranial indices close to normal. The cranial index is the most widely used aesthetic end point in the literature, and while it has many shortcomings as an outcome measurement, its appeal is its broad usage. Small differences in cranial indices between procedures may, or may not, correlate with a significant difference in appearance, and this unknown measure is a limitation of our study and others. Age was also significantly older for patients undergoing pi-plasty due to the increased blood loss and quality of bone in younger patients.

Cranial vault remodeling was first described in 1996[5] by the same group performing the comparison with SMC. Initially, they published a small series of SMC patients,[4] but later reported on 75 SMC patients, the largest series to date.[3] This study commented on 3- and 5-year outcomes of patients undergoing SMC and found that the cranial index remained normalized after spring removal. Even when including the surgery for spring removal, SMC patients had significantly less blood loss, blood transfusions, operative time, ICU time, hospital days, and overall costs than pa-

TABLE 3. Overview of studies directly comparing outcomes of SMC to other procedures

Procedure Comparison	Authors & Year	No. of Patients	Age (mos)	Blood Loss (ml)	Surgery Time (min)	Postop Stay (days)	ICU Stay (hrs)	Preop CI	Postop CI
SMC vs pi-plasty									
	Windh et al., 2008								
	SMC	20	3.5	170	104	6	25	66.57	70.57
	Pi-plasty	20	7.1	425	121	9	46	66.12	73.37
	p value		<0.001	<0.001	0.0103	<0.001	0.03	0.9676	0.0128
	Guimarães-Ferreira et al., 2003								
	SMC	10	3.2	60.3 (% ERCM)	100	6.4	23.9	65.15	71.23
	Pi-plasty	10	5.8	94.5 (% ERCM)	110	8.6	25.7	66.24	72.65
	p value		0.002	0.003	0.464	0.013	0.619	0.331	0.276
SMC vs CVR									
	David et al., 2010								
	SMC	75	5.7	25	30	0.93	0	69	75.4
	CVR	18	19	255	143	>4	18	66	72.5
	p value		<0.001	<0.001	<0.001	<0.001	<0.001	0.036	
SMC vs SC									
	Mackenzie et al., 2009								
	SMC	9	7	9.6 (% EBV)	66	4.4			
	SC	13	10	17 (% EBV)	76	7			
SMC vs SCPB									
	Taylor et al., 2011								
	SMC	7	3.7	24.3	46	1.4	16.8	64.9	78.9
	SCPB	7	2.3	90.7	115.4	2.3	24	65.3	78
	p value		<0.001	<0.001	0.002	0.009	0.147	0.862	0.643
	Mackenzie et al., 2009								
	SMC	9	7	9.6 (% EBV)	66	4.4			
	SCPB	5	7	16 (% EBV)	86	6			
SMC vs SCBCS									
	Mackenzie et al., 2009								
	SMC	9	7	9.6 (% EBV)	66	4.4			
	SCBCS	7	10	14 (% EBV)	122	5			

EBV = estimated blood volume; ERCM = estimated red cell mass (substituted); SC = strip craniectomy; SCBCS = strip craniectomy with bioabsorbable cross-struts; SCPB = strip craniectomy with parietal barrel staving.

tients undergoing CVR.[3] Additionally, age again was significantly older in CVR due to the morbidity of undergoing extensive surgery at a younger age.

Minimally invasive techniques including strip craniectomy with and without parietal barrel staving and strip craniectomy with bioabsorbable plates have been less extensively compared with SMC, but the minimum equipoise among these techniques has been achieved. It was observed that patients with parietal barrel stavings had longer hospital stays, operative time, and estimated blood loss when compared with SMC.[22] These similar results are not unexpected, however, because SMC is basically a variation on the strip craniectomy technique that provides parents the option to circumvent the constraints of wearing a helmet for several months.

Although the literature review has demonstrated the benefits of SMC, there are several issues with the current reported literature. The number of institutions contributing to the SMC literature is minimal, with most of the reports coming from Sahlgrenska University Hospital, Göteborg, Sweden,[8,16,27] and Wake Forest University, Winston Salem, North Carolina, with a few exceptions of single papers from other institutions.[22,25] One major advantage of any of the other techniques, including endoscopic strip craniectomy with helmet remodeling, over SMC is that a second procedure is not normally needed. In our study we have included the outcomes for the second operation. Most studies in the literature, including those in our pooled data set, do not include these measurements. However, even when accounting for both surgeries, the patients at CHOP demonstrated a lower total blood loss and length of postoperative hospital stay than the pooled patient data when compared with other SMC studies. Although direct comparisons cannot be drawn between our study and cranial vault reconstruction, David et al.[3] has shown improved outcomes in SMC patients over CVR patients and our SMC data are similar for length of postoperative stay and estimated blood loss. At CHOP, most children stay for observation until postoperative Day 1 for both the spring placement and the removal procedures. This stay allows for adequate pain control but may add to the cost of the procedure. The cost of this additional stay is difficult to assess as costs are so variable at different institutions, but we estimate the cost of our operation, even with the longer ICU stay than other SMC surgeries, to be well under the cost of a cranial vault surgery. David et al.[3] estimated the cost of SMC with removal of the springs to be $12,766, while the cranial expansion cost was $27,212. This cost comparison should closely match the cost of our procedure as our length of stay was very similar to that in the study of David et al.,[3] with the exception of 1 day of ICU stay. The blood loss was minimal and the second surgeries were performed well after standard recuperation time from the spring placement. It will be interesting to see whether technological advances in biomaterials will allow for resorbable springs, thus eliminating the need for an additional surgery.

Due to the relative novelty of the spring-mediated technique, the surgery has evolved over time, making comparisons between papers difficult. When first reported, a single spring with large toothed footplates was placed through a large S-shaped incision to achieve suture expansion.[15] In this paper we report modifications to this technique using multiple springs placed through 2 small incisions. Although results from the studies are similar, pooling data is difficult due to minor differences in technique, and these inconsistencies could skew results. A more standardized clinical trial with multiple institutions may be able to discern the true advantages of varying SMC techniques.

Furthermore, outcome measures were not truly uniform across all papers and are at times evasive. At CHOP, radiation minimization is the goal when evaluating patients preoperatively and postoperatively. Patients routinely get a preoperative 3D CT scan to evaluate for multiple suture synostosis. Postoperative patients receive a cephalogram to assess for spring placement. These two studies of differing radiological techniques, while clinically relevant, make the calculation of cranial index difference difficult. This discrepancy could have inadvertently skewed our results, although both are validated methods for cranial index calculation.[24] Additionally, many of the SMC studies did not report a method for cranial index measurements.[3,17,22,25] Of the studies that did clarify these methods, acquisition of cranial index calculations varied, including the use of cephalograms[25,27] and laser Doppler ultrasonography.[3] This variation makes pooling data from multiple studies difficult because very little evaluation has been performed to compare methods from different modalities. One recent study contends that 3D CT cranial index measurements are closer to digital caliper measurements of cranial index than are 2D CT measurments.[24] A standard method for measuring the cranial index would help with directly comparing the results from multiple institutions. Noninvasive techniques that are reproducible would be ideal, but this option should be explored further. Subjective measures show that patients were satisfied with SMC corrections, but this outcome measure cannot be easily compared as none of our patients have had to undergo correction surgery, and all families have been satisfied with outcomes. Future evaluations should include a standard follow-up measurement strategy that can be easily compared among multiple institutions.

Lastly, the literature for SMC is centered on comparing cosmetic results and evaluating the morbidity of the procedure. This focus on cosmetic outcomes has diverted attention from the cognitive changes that occur after sagittal synostosis surgery. A multiinstitutional evaluation[9] of the minimally invasive endoscopic strip craniectomy compared with cranial vault repair found the latter to have better long-term cognitive testing outcomes despite the increased morbidity and later age of operation. This brings up several important considerations when treating sagittal craniosynostosis.

First, it remains unknown whether the results of such a study on endoscopic strip craniectomy can be extrapolated to SMC. A major difference between strip craniectomy and SMC is the need for an extended time in an external orthosis following a strip-only technique. The effect on neurodevelopment of short-term use of an external orthotic helmet in the setting of sagittal synostosis has not been evaluated by us, or by anyone else in the literature, and thus it would be impossible to comment on whether it may have a negative or positive effect. Although some of our patients received a short-term orthosis for up to 2 months preoperatively, it is unlikely that this would affect cognitive development as much as the lengthy period during critical cognitive development that occurs with the endoscopic strip craniectomy.[23] In addition, SMC is a distracting device that may allow skull expansion over time as the brain grows.

Second, the extent to which a potential cognitive difference should outweigh the positive attributes of SMC remains difficult to discern. The cognitive consequences of surgical procedures are critical to consider and future clinical studies examining the cognitive effects of SMC on patients will provide important feedback for future surgical interventions.

Conclusions

Our study corroborates findings in the literature for the efficacy of SMC in the treatment of sagittal craniosynostosis. We have been able to use techniques at the CHOP that further reduced morbidity compared with other SMC procedures. Spring-mediated cranioplasty has been shown to produce similar outcomes as more invasive procedures while reducing harm to patients and facilitating surgical efficiency. Our clinical data will contribute to the growing literature on SMC across different institutions. Future studies examining the overall effects of such surgical interventions on cognitive function for patients with sagittal craniosynostosis will help elucidate whether SMC is safe and favorable in both the short- and long-term compared with other more invasive forms of CVR.

References

1. Albright AL: Operative normalization of skull shape in sagittal synostosis. **Neurosurgery 17:**329–331, 1985
2. Chaichana KL, Jallo GI, Dorafshar AH, Ahn ES: Novel use of an ultrasonic bone-cutting device for endoscopic-assisted craniosynostosis surgery. **Childs Nerv Syst** [epub ahead of print], 2013
3. David LR, Plikaitis CM, Couture D, Glazier SS, Argenta LC: Outcome analysis of our first 75 spring-assisted surgeries for scaphocephaly. **J Craniofac Surg 21:**3–9, 2010
4. David LR, Proffer P, Hurst WJ, Glazier S, Argenta LC: Spring-mediated cranial reshaping for craniosynostosis. **J Craniofac Surg 15:**810–818, 2004
5. David LR, Taylor RG, Argenta LC: A technique for cranial bone expansion. **J Craniofac Surg 7:**151–155, 1996
6. David LR, Wilson JA, Watson NE, Argenta LC: Cerebral perfusion defects secondary to simple craniosynostosis. **J Craniofac Surg 7:**177–185, 1996
7. Epstein N, Epstein F, Newman G: Total vertex craniectomy for the treatment of scaphocephaly. **Childs Brain 9:**309–316, 1982
8. Guimarães-Ferreira J, Gewalli F, David L, Olsson R, Friede H, Lauritzen CG: Spring-mediated cranioplasty compared with the modified pi-plasty for sagittal synostosis. **Scand J Plast Reconstr Surg Hand Surg 37:**208–215, 2003
9. Hashim PW, Patel A, Yang JF, Travieso R, Terner J, Losee JE, et al: The effects of whole-vault cranioplasty versus strip craniectomy on long-term neuropsychological outcomes in sagittal craniosynostosis. **Plast Reconstr Surg 134:**491–501, 2014
10. Hunter AG, Rudd NL: Craniosynostosis. I. Sagittal synostosis: its genetics and associated clinical findings in 214 patients who lacked involvement of the coronal suture(s). **Teratology 14:**185–193, 1976
11. Jane JA, Edgerton MT, Futrell JW, Park TS: Immediate correction of sagittal synostosis. **J Neurosurg 49:**705–710, 1978
12. Jimenez DF, Barone CM: Endoscopic craniectomy for early surgical correction of sagittal craniosynostosis. **J Neurosurg 88:**77–81, 1998
13. Lane L: Pioneer craniectomy for relief of mental imbecility due to premature sutural closure and microcephalus. **JAMA 18:**49–50, 1892
14. Lannelongue M: De la craniectomie dans la microcephalie. **Compt Rend Seances Acad Sci 50:**1382–1385, 1890
15. Lauritzen C, Sugawara Y, Kocabalkan O, Olsson R: Spring mediated dynamic craniofacial reshaping. Case report. **Scand J Plast Reconstr Surg Hand Surg 32:**331–338, 1998
16. Lauritzen CG, Davis C, Ivarsson A, Sanger C, Hewitt TD: The evolving role of springs in craniofacial surgery: the first 100 clinical cases. **Plast Reconstr Surg 121:**545–554, 2008
17. Mackenzie KA, Davis C, Yang A, MacFarlane MR: Evolution of surgery for sagittal synostosis: the role of new technologies. **J Craniofac Surg 20:**129–133, 2009
18. Sgouros S, Goldin JH, Hockley AD, Wake MJ, Natarajan K: Intracranial volume change in childhood. **J Neurosurg 91:**610–616, 1999
19. Shillito J Jr, Matson DD: Craniosynostosis: a review of 519 surgical patients. **Pediatrics 41:**829–853, 1968
20. Sood S, Rozzelle A, Shaqiri B, Sood N, Ham SD: Effect of molding helmet on head shape in nonsurgically treated sagittal craniosynostosis. **J Neurosurg Pediatr 7:**627–632, 2011
21. Tamburrini G, Caldarelli M, Massimi L, Santini P, Di Rocco C: Intracranial pressure monitoring in children with single suture and complex craniosynostosis: a review. **Childs Nerv Syst 21:**913–921, 2005
22. Taylor JA, Maugans TA: Comparison of spring-mediated cranioplasty to minimally invasive strip craniectomy and barrel staving for early treatment of sagittal craniosynostosis. **J Craniofac Surg 22:**1225–1229, 2011
23. Thompson RA, Nelson CA: Developmental science and the media. Early brain development. **Am Psychol 56:**5–15, 2001
24. van Lindert EJ, Siepel FJ, Delye H, Ettema AM, Bergé SJ, Maal TJ, et al: Validation of cephalic index measurements in scaphocephaly. **Childs Nerv Syst 29:**1007–1014, 2013
25. van Veelen ML, Mathijssen IM: Spring-assisted correction of sagittal suture synostosis. **Childs Nerv Syst 28:**1347–1351, 2012
26. Venes JL, Sayers MP: Sagittal synostectomy. Technical note. **J Neurosurg 44:**390–392, 1976
27. Windh P, Davis C, Sanger C, Sahlin P, Lauritzen C: Spring-assisted cranioplasty vs pi-plasty for sagittal synostosis—a long term follow-up study. **J Craniofac Surg 19:**59–64, 2008

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