Spring-mediated Cranioplasty Versus Minimally Invasive Endoscopic Craniosynostosis Repair In Young Infants:
A Multicenter Study from the Pediatric Craniofacial Collaborative Group

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Background: Endoscopic strip craniectomy (ESC) and spring-mediated cranioplasty (SMC) are two less invasive surgical techniques for treating craniosynostosis in early infancy. The treatment algorithm of ESC requires postoperative helmet therapy whereas SMC requires strip craniectomy with insertion of 2-3 spring expansion devices and a second surgery for spring removal 2-3 months later. In this report from the pediatric craniofacial collaborative group (PCCG) we hypothesized that perioperative outcome measures were similar between SMC-insertion versus ESC using a multicenter database.

Methods: After local IRB approval 33 institutions contributed data of 676 infants from June 2012 to September 2019. We analyzed 580 infants (<6 months) from 32 centers undergoing ESC and 96 infants from 5 centers undergoing SMC (strip craniectomy with insertion of springs only). Primary outcomes included transfusion-free hospital course, ICU utilization and hospital length of stay (HLOS); secondary outcomes included perioperative blood transfusion, blood donor exposures, length of surgery and intraoperative use of vasopressors. We also obtained data points for SMC-spring removal. SMC patients were 1:2 propensity score (PS) matched on age (months) and weight (kg) with ESC patients using a greedy matching algorithm. Comparison of outcomes between PS matched groups was performed with descriptive analyses and multivariable regression models.

Results: Demographic data are shown in Table 1. After matching 96 SMC to 192 ESC patients, patient characteristics (except ASA class and antifibrinolytics) were balanced for both groups (Table 1). Table 2 reports descriptive analyses of outcomes between PS matched groups. Multivariable regression models adjusted for ASA class and antifibrinolytics (Table 3) showed no difference in transfusion- free hospital course (P=0.3774). However patients with SMC-spring insertion were more likely to be admitted to the ICU (OR: 7.50, 95% CI 3.75-14.99, P<0.0001), had longer ICU LOS (IRR: 1.42, 95% CI, 1.37-1.48, P<0.0001), and HLOS (IRR:1.28, 95%CI, 1.17-1.39, P<0.0001). Secondary outcomes were not significantly different between these groups. For the second surgical procedure of SMC-spring removal (N=91) the median surgical duration time was 33 min (IQR, 26-42), one infant required blood transfusion, one infant was admitted to the ICU and 33 infants were hospitalized (Table 4, 5).

Discussion and Conclusion: Infants with SMC-insertion had significantly higher ICU admission rates, longer ICU stay and HLOS compared to ESC however surgical duration & transfusion outcomes were similar between groups. The effect of a second surgical/anesthetic exposure of SMC vs helmet therapy of ESC on the developing brain in young infants needs to be determined.

Table 1: Patient Characteristics: SMC-Spring Insertion Versus Endoscopic Strip Craniectomy (ESC) Before and After Propensity Score Matching

	BEFORE PROPENSITY SCORE MATCHING			AFTER PROPENSITY SCORE MATCHING		
Characteristics	SMC (N = 96)	ESC (N = 580)	P-value	SMC (N = 96)	ESC (N = 192)	P-value
Age (months), Median (Q1, Q3)	3.0 (3.0, 3.0)	3.0 (2.0, 3.0)	<.0001*	3.0 (3.0, 3.0)	3.0 (3.0, 3.0)	0.9999
Weight (kg), Median (Q1, Q3)	6.2 (5.7, 6.9)	6.0 (5.5, 6.8)	0.0744	6.2 (5.7, 6.9)	6.2 (5.7, 6.9)	0.4895
Gender, n (%)			0.6297			0.2483
Male	75 (78.1)	440 (75.9)		75 (78.1)	142 (74.0)	
Female	21 (21.9)	140 (24.1)		21 (21.9)	50 (26.0)	
Race, n (%)			0.4475			0.2254
White	76 (79.2)	444 (76.6)		76 (79.2)	145 (75.5)	
Black	4 (4.2)	19 (3.3)		4 (4.2)	4 (2.1)	
Asian	1 (1.0)	13 (2.2)		1 (1.0)	1 (0.5)	
Other	14 (14.6)	76 (13.1)		14 (14.6)	32 (16.7)	
Not recorded/Unknown	1 (1.0)	28 (4.8)		1 (1.0)	10 (5.2)	
ASA class, n (%)			<.0001*			0.0074*
1/2	71 (74.7)	514 (89.2)		71 (74.7)	168 (88.0)	
3	24 (25.3)	62 (10.8)		24 (25.3)	23 (12.0)	
Antifibrinolytic, n (%)	56 (58.3)	166 (29.1)	<.0001*	56 (58.3)	63 (33.5)	0.0006*
Preoperative, Median (Q1, Q3)						
Hgb (g/dL)	11.1 (10.5, 11.8)	11.1 (10.4, 11.8)	0.9653	11.1 (10.5, 11.8)	11.1 (10.4, 11.8)	0.1681
HCT (%)	32.8 (30.8, 35.3)	32.6 (30.2, 34.5)	0.3220	32.8 (30.8, 35.3)	32.6 (30.2, 34.2)	0.2085
Platelet Count (10^9/L)	449.0 (385.0,517.5)	450.0 (388.0,519.0)	0.5689	449.0 (385.0, 517.5)	453.0 (391.0, 516.0)	0.3055
Diagnosis category, n (%)						
Sagittal	96 (100.0)	580 (100.0)		96 (100.0)	192 (100.0)	

ASA class, American Society of Anesthesiology Classification; ESC, endoscopic strip craniectomy; HCT, hematocrit; Hgb, hemoglobin; PSM, propensity score matching; SMC, spring-mediated cranioplasty.
Age, weight and laboratory values are median and interquartile range
Before PSM: Patient data captured from 33 distinct centers. SMC - 5 centers; ESC - 32 centers.
After PSM: Patient data captured from 29 distinct centers. SMC - 5 centers; ESC - 26 centers.

Table 2. Comparison of Outcomes Between SMC-Spring Insertion and Endoscopic Strip Craniectomy: Propensity Score Matched Groups (N = 288)

Outcomes	SMC (N = 96)	ESC (N = 192)	P-value
Primary			
Transfusion-free hospital course, n (%)	56 (58.3%)	126 (65.6%)	0.2272
ICU admission, n (%)	83 (86.5%)	67 (35.0%)	<.0001*
ICU length of stay, (SMC [n=94], ESC [n=179])	2.0 (2.0, 2.0)	0.0 (0.0, 2.0)	<.0001*
Hospital length of stay, (SMC [n=95], ESC [n=186])	3.0 (2.0, 3.0)	2.0 (2.0, 3.0)	<.0001*
Secondary			
Perioperative RBC product transfusion (mL/kg)	0.0 (0.0, 15.1)	0.0 (0.0, 14.9)	0.2176
Total perioperative blood donor exposures	0.0 (0.0, 1.0)	0.0 (0.0, 1.0)	0.2641
Duration of surgery (min), (SMC [n=96], ESC [n=190])	79.0 (70.0, 94.5)	75.5 (52.0, 102.0)	0.1073
Intraoperative IV epinephrine bolus, n (%), (SMC [n=96], ESC [n=190])	4 (4.2%)	2 (1.1%)	0.0798
Intraoperative vasopressor infusion, n (%), (SMC [n=96], ESC [n=189])	0 (0.0)	6 (3.2%)	0.1005

ESC, endoscopic strip craniectomy; ICU, intensive care unit; RBC, red blood cell; SMC, spring mediated cranioplasty.
Continuous data are median (IQR); Categortical data are n (%). Groups are compared with descriptive analyses.
* Statistically significant group differences.

Table 3: Multivariable Analysis Comparing Outcomes Between SMC-Spring Insertion and ESC Matched Groups (N = 288)

Outcomes	Estimates	P-value
Primary		
Transfusion-free hospital course^		
aOR (95% CI)	0.78 (0.45-1.35)	0.3774
ICU admission^		
aOR (95% CI)	7.50 (3.75-14.99)	<.0001*
ICU length of stay^^^		
IRR (95% CI)	1.42 (1.37-1.48)	<.0001*
Hospital length of stay^^^^		
IRR (95% CI)	1.28 (1.17-1.39)	<.0001*
Secondary		
Periop RBC product transfusion (mL/kg)^^^^^		
β (95% CI)	0.73 (-1.96-3.42)	0.5948
Total perioperative blood donor exposures^^		
OR (95% CI)	1.26 (0.75-2.11)	0.3825
Duration of surgery (minutes)^^^^^		
β (95% CI)	1.12 (-7.11-9.35)	0.6395

aOR, adjusted odds ratio; IRR, incidence risk ratios; β, beta estimate; CI, confidence interval; ICU, intensive care unit; RBC, red blood cell.
Models adjusted for ASA class and antifibrinolytic administration.
^Results from a conditional logistic regression model. ^^Results from an ordinal logistic regression model.
^^^Results from a zero-inflated Poisson regression model. ^^^^Results from a Poisson regression model.
^^^^^Results from a linear regression model.

Table 4: Perioperative Outcomes for SMC-Spring Removal Surgery

Outcomes	N (%) or Median (IQR)
Transfusion-free hospital course	90 (99)
Day surgery	62 (64)
Day surgery length of stay (hours)	6 (5-8)
Ward admission	33 (35)
Ward admission length of stay (days [b])	2 (2-2)
ICU admission	1 (1)
ICU admission length of stay (days [b])	3 (3-3)
Duration of surgery (minutes)	33 (26-42)

[a] Spring removal surgery data was available from 1 center: 91 of 96 cases from spring insertion dataset.
[b] Length of stay data reported as median (IQR). Ward and ICU length of stay data includes partial or whole calendar days.

Table 5: Comparison of SMC-Spring Insertion, Spring Removal, and ESC

Outcomes	SMC insertion (N=96)	ESC (N=192)	Spring removal (N=91)
Transfusion-free hospital course, n (%)	56 (58.3)	126 (65.6)	90 (99)
ICU admission, n (%)	83 (86.5)	67 (35.0)	1 (1)
Duration of surgery (minutes), Median (IQR)	79 (70-95)	76 (52-102)	33 (26-42)