Published in final edited form as:

J Craniofac Surg. 2013 July ; 24(4): 1331–1335. doi:10.1097/SCS.0b013e3182997885.

Laypersons' ratings of appearance in children with and without single suture craniosynostosis

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Abstract

Background—Single suture craniosynostosis (SSC) results in head shape anomalies that likely affect social perceptions of appearance. The purpose of this study was to evaluate laypersons' ratings of attractiveness in children with and without SSC. Among cases, we also examined differences by suture fused and age at surgery.

Methods—We collected photographs of 196 children with SSC and 186 children without SSC as infants (prior to surgery, for cases) and at ages 18 and 36 months. Photographs were rated by 8 raters, who were blinded to the population being studied. We used linear regression to compare appearance ratings for the two groups at each visit and to evaluate changes over time. Regression analyses were used to examine the association between age at surgery and appearance ratings.

Results—Children with SSC received lower appearance ratings than unaffected controls at each visit (all p-values < .001). Appearance ratings decreased over time, with a similar trajectory for children with and without SSC. Among cases, those with unicoronal and lambdoid synostosis had

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Conflict of Interest:

The authors have no conflicts of interest to report.

the lowest ratings and those with sagittal synostosis had the highest. Age at surgery was inversely associated with appearance ratings.

Conclusions—Children with SSC received lower appearance ratings than unaffected controls, with minimal change following surgery. Better outcomes were associated with earlier surgery. These findings do not indicate that children with SSC failed to benefit from surgery, as without surgical intervention, asymmetrical head shape would likely have worsened over time. However, our data suggest that appearance does not fully ‘normalize’.

Keywords

Infant; Craniosynostosis; Appearance

INTRODUCTION

Single suture craniosynostosis (SSC) is a birth defect involving the premature fusion of one of the 6 major cranial sutures (metopic, sagittal, right or left coronal, or right or left lambdoid). The overall prevalence of SSC is approximately 1 in 2,000 live births.¹⁻⁴ Children born with SSC typically receive cranioplasty to release the fused suture and reshape the deformed calvaria, with the primary goals of reducing intracranial pressure and normalizing head shape and facial appearance.⁵ Surgical outcome studies for SSC have relied on measures of head shape as the primary outcome of interest. For example, using the cephalic index and similar measures of irregularity in head shape, several authors have shown improvements following cranioplasty and other procedures.⁷⁻¹⁰ Surgeon's impressions or ratings of specific cranial features pre and post-surgery have also been used to document outcomes, and have generally shown improvement following surgery.¹¹

Although head shape indices and surgeons' ratings can offer precise measurement of outcome, the relation between these measures and the social perceptions of peers and adult caregivers is unclear. In children with craniofacial anomalies, ratings by lay observers (i.e., untrained peers or adults, who are unaware of the condition being studied) potentially provide a “real world” assessment of how noticeable a child's condition is in day-to-day interactions. There is an extensive literature, primarily with non-craniofacial populations, showing that perceived appearance is associated with self concept, social acceptance, and teachers' expectations and evaluations.⁶

In conjunction with an ongoing study of neurodevelopment, we sought to evaluate laypersons' ratings of appearance for children with and without SSC. We used procedures adapted from the social psychology literature to capture impressions based on front-view facial photographs taken when children were infants (Time 1; for cases, prior to surgery) and again at ages 18 and 36 months (Time 2 and Time 3, respectively). This was not intended as a surgical outcome study. However, we were interested in documenting changes in appearance for children with and without SSC over time to determine whether any group differences persisted after cranioplastic surgery and as children physically matured. We also examined the relation between timing of surgery and appearance outcomes. Surgery for children with SSC is typically performed within the first year of life, to capitalize on the infant's more malleable calvarium and to allow the cranial shape to normalize with brain growth.⁵ However, findings are mixed on the association between age at surgery and outcomes in head shape or cosmesis. Although some studies show better outcomes with earlier surgery,^{12,13} others have indicated either no association between outcome and age at surgery⁹ or worse outcomes in younger infants.¹⁴

MATERIALS AND METHODS

Sample

All participants were recruited through the “Infant Learning Project,” a multi-site, longitudinal cohort study of development in children with SSC (See Speltz et al.¹⁵). Participants were enrolled after informed consent had been obtained from their parents. The study was approved by the institutional review boards (IRB) from each participating center: Seattle Children's Hospital; Children's Memorial Hospital and Northwestern University in Chicago; Children's Healthcare of Atlanta, St. Louis Children's Hospital; University of Illinois in Chicago (UIC), the University of Chicago (UC) and Children's Hospital of Philadelphia (CHOP).

Patients

Infants with SSC were referred to the project at the time of diagnosis by the treating surgeon or pediatrician. Infants were eligible if they: (1) had sagittal, metopic, unilateral coronal, or unilateral lambdoid craniosynostosis confirmed by computed tomography scans; (2) had yet to undergo cranial vault surgery and (3) were 30 months of age or younger at the time of recruitment. Infants were excluded for: 1) prematurity (<34 weeks gestation); 2) presence of major medical or neurological conditions (for example, cardiac defects, seizure disorders, cerebral palsy, or other significant health conditions requiring major surgery); 3) presence of three or more extracranial minor malformations as defined by Leppig et al.¹⁶ or 4) presence of major malformations. Twins were eligible to participate in the study if one of them had SSC (in the event of twins who both had SSC, one was randomly selected for participation). A total of 270 patients were enrolled in the parent study, representing 84% of all those eligible. Two hundred forty patients participated at Time 2 (89%) and 210 participated at Time 3 (78%).

Controls

Infants in the control group had no known craniofacial condition and did not meet any of the exclusionary criteria for patients described above. We recruited control infants who were generally similar to patients, taking into consideration: 1) age, 2) sex, 3) family socioeconomic status,¹⁹ and 4) ethnicity. A pool of 581 control group participants was recruited through pediatric practices, birthing centers and announcements in newsletters and/or other publications of interest to parents of newborns. The 259 originally enrolled control group participants represented 44% of those who were eligible. Among controls who were not enrolled, most (76%) were infants who “aged out” of the project before they could be matched to an enrolled case. Two hundred forty controls participated at Time 2 and 224 participated at Time 3 (93% and 86% of those seen at Time 1, respectively).

Procedures

Cases' Time 1 visits were scheduled prior to surgery, typically when children were < 12 months old. Target ages for the Time 2 and Time 3 visits were 18 and 36 months, respectively. Thirty patients received surgery between 12–24 months, and post-surgery visits for these patients were scheduled to ensure an interval of 6 months at Time 2. A few patients (n=5) received surgery after 24 months, and completed only a single post-surgery assessment. Controls were frequency matched to the case group for age at Time 1.

Multiple front-view digital photographs were taken at each of the study visits. Photographs were selected for rating if they provided a clear view of the child's face with a neutral expression. Digital photographs were cropped to eliminate background and the child's clothes, which may bias attractiveness ratings. Sample photographs are presented in Figure 1.

Eight adult female raters were recruited from area daycare centers, ensuring adequate familiarity with young children. Raters were diverse in terms of ethnicity (n = 4 Caucasian, n=2 Asian, and n=2 mixed ethnicity or other) and age (median age = 38.5 years, SD = 12.3). Raters had a median of 7 years (SD = 8.1) of childcare experience. Raters were told that the study focused on facial attractiveness in young children. However, they were not informed that some participants had a craniofacial anomaly or that some participants underwent surgery. Raters completed their review of Time 1 photographs for cases and controls before proceeding to Time 2 photographs for both groups. Six of the 8 raters returned later to complete a review of Time 3 photographs.

Photographs were presented, in randomized order, on a desktop computer screen. Raters were asked to place the photographs in one of seven folders corresponding to the child's attractiveness, ranging from 1 = "Highly Unattractive" to 7 = "Highly Attractive." Raters were encouraged to periodically review their selections and to make adjustments as needed. For example, after reviewing the first few photographs, raters were encouraged to assess their initial ratings to determine whether they were too high or too low in relation to their subsequent judgments and, if so, to adjust their ratings. Inter-rater agreement was relatively low at each time point, ranging from intra-class correlation (ICC) = 0.14 to 0.22 for cases and ICC = 0.14 to 0.17 for controls. For analyses, attractiveness scores for the 8 raters (6 raters at Time 3) were averaged, resulting in a single attractiveness score at each time point.

Analyses

Descriptive statistics (means, standard deviations) were used to summarize average attractiveness ratings separately for cases and controls at each of the three visits. We used a generalized estimating equations (GEE) approach to linear regression with robust standard error estimates to compare average attractiveness ratings between cases and controls at each time point, adjusting for child's age at the visit, sex, race/ethnicity (White, Non-Hispanic; Non-White or Hispanic), and study site. We included visit number in the model as indicator variables (two indicators) and also included an interaction between case status and these indicators, which allowed for analysis of differences in 1) slope from Time 1–2 and Time 2–3, and 2) the trajectories of cases and controls. Using the same model we also examined the change in average attractiveness rating from Time 1 to Time 2 and Time 2 to Time 3 within cases and controls separately. We performed a joint Wald test of the two interaction terms to test whether case-control group differences changed with time. An advantage of GEE relative to other analytic strategies (e.g., analysis of variance) is that GEE retains subjects who have missing data at one or more visits. This allowed us to include all participants with attractiveness ratings at one or more study visits, and reduces bias due to missing data. Nonetheless, to ensure that findings remained unchanged, we ran sensitivity analyses restricted to those participants who were seen for all visits and to raters who provided attractiveness ratings at each time point. We also explored differences in attractiveness as a function of diagnosis (i.e., location of suture fusion) by summarizing the mean and standard deviation of average attractiveness rating at each time point within each suture group and in controls. Using a t-test with unequal variances, we compared the means of each suture group to that of the control group at the three time points. Because of the limited sample size within each suture group (particularly the lambdoid group), we were unable to adjust for any confounders.

Finally, among cases we examined the association between age of surgery and post-surgery facial attractiveness using linear regression with robust standard error estimates. Attractiveness ratings at Time 2 and Time 3 were evaluated in separate regressions as a function of age at surgery, adjusting for pre-surgery attractiveness rating, age at the visit, sex, race/ethnicity, and study site. Because age at surgery varies depending on the affected

suture, we further adjusted these analyses by suture (sagittal, metopic, unicoronal, or lambdoid).

Analyses were completed using STATA 12 software (Ref).

RESULTS

At least one photograph was available for 196 children with SSC (“cases”) and 186 children without SSC (“controls”). Seventy five controls and 94 cases had photographs for each of the three visits. Photographs were unavailable for 42 controls and 32 cases who participated in follow-up visits, but either did not have a photograph taken or had a photograph of poor quality (e.g., poor focus, inadequate view of the child’s face, non-neutral expression). Participants with useable photographs were similar to those without photographs in terms of age (7.4 months vs. 7.5 months, respectively) and gender (52% male vs. 48% male, respectively). Those without photographs were more often controls (60% vs. 41% participants with photos) and non-Caucasian (58% vs. 47% participants with photos). We excluded from all analyses 3 cases who had not received surgery by their Time 3 visit. Another case participant, who received surgery after the Time 2 visit, was excluded from Time 2 analyses but retained for all other analyses.

Descriptive statistics for participating cases and controls are summarized in Table 1. At Time 1 the mean age was 7.6 months in controls and 7.3 months in cases; most case and controls subjects were male, white, of high SES, and recruited from the Seattle site. These characteristics were similar in cases and controls at Times 2 and 3. Among cases, mean age at surgery varied as a function of affected suture, ranging from 7.1 months for children with sagittal synostosis to 11.1 months for children with unicoronal synostosis (Table 2).

The unadjusted mean attractiveness rating for controls ranged from 4.6 to 5.0 and from 4.0 to 4.4 for cases across the visits (Table 3). In GEE analyses, cases had lower estimated attractiveness scores than controls at each time point (Time 1: -0.71 , 95% CI -0.88 , -0.54 ; Time 2: -0.55 , 95% CI -0.72 , -0.38 ; Time 3: -0.58 , 95% CI -0.74 , -0.41) (Table 4). Among cases, there was almost no change in attractiveness rating from Time 1 to Time 2 (-0.02 , 95% CI -0.23 , 0.19), but a decrease of 0.84 points from Time 2 to Time 3 (95% CI -1.15 , -0.52). In controls there was a similar pattern with a small decrease between Time 1 and Time 2 (-0.17 , 95% CI -0.38 , 0.04) and a larger decrease between Time 2 and Time 3 (-0.81 , 95% CI -1.11 , -0.51). There was little evidence for change over time in case-control group differences, indicating a similar trajectory for the two groups (p -value for Wald test= 0.15) (Table 4). These findings remained unchanged when restricted to participants with data from all three study visits (results not shown). Similarly, there was minimal change when we restricted analyses to the 6 raters who completed attractiveness ratings at all three time points (results not shown).

Across time, each of the diagnostic groups had lower mean attractiveness ratings than the control group, with the unicoronal and lambdoid groups having the lowest mean rating at all time points and the sagittal group having the highest mean rating (Table 2). In children with sagittal synostosis the difference compared to controls ranged from -0.29 to -0.62 points (p 0.007 for all), -0.67 to -0.71 in children with metopic synostosis (p < 0.001 for all), -0.75 to -0.91 in children with unicoronal synostosis (p < 0.001 for all), and -0.74 to -0.90 in children with lambdoid synostosis (p = 0.02 to 0.21).

When examining the association between age at surgery and post-surgery facial attractiveness, the estimated average attractiveness rating at Time 2 decreased by 0.16 points for every six-month increase in the age at surgery (95% CI -0.34 , 0.00); at Time 3

attractiveness rating decreased by 0.25 points for every six-month increase (95% CI -0.42 , -0.07).

DISCUSSION

To our knowledge, our study is the first to examine longitudinal trends in aesthetic outcome for children with SSC versus unaffected controls, using ratings by naïve observers. In contrast to previous studies with this population, which have utilized measures of head shape and surgeons' ratings to document changes following surgery, our intent was to capture a 'real world' measure of perceived appearance. As has been observed in studies of children without craniofacial anomalies,^{17,18} appearance ratings in both groups declined from infancy to age 36 months. Children with SSC were consistently rated as less attractive than unaffected controls by naïve raters, with minimal change in group differences between infancy and age 36 months.

Our investigation was not intended as a surgical outcome study, and the findings should not be taken to suggest that children failed to derive benefit from surgery. Without surgery, deformities associated with SSC are likely to become more apparent over time as the brain continues to grow and the skull becomes increasingly asymmetric. In the United States, it is rare for children diagnosed with SSC not to undergo cranial vault surgery, making it difficult to quantify outcomes for children who receive surgery versus those who do not. Further, we did not assess children's appearance from multiple perspectives (e.g., profile, top down), and post-surgery changes may be more readily observed from these alternative views. Nonetheless, our findings do not indicate that appearance fully 'normalizes' for children with SSC by age 36 months.

Case-control group differences were present for all SSC diagnoses, though tended to be largest for cases with unicoronal and lambdoid synostosis, a likely reflection of the asymmetric deformities associated with these fusions as opposed to the more symmetric presentation of sagittal and metopic synostosis. Age of surgery was inversely associated with cosmetic outcomes, even after adjusting for site of suture fusion. This finding might suggest that, in general, outcomes are better for children who received surgery at a younger age. However, we are unable to control for other potential confounds that might account for this association. For example, in addition to the location of suture fusion, age at surgery may vary as a function of age at diagnosis or severity of deformation. It should also be noted that other factors have to be considered when determining the timing of surgery (e.g., type of procedure, intracranial pressure), which may make it impractical or undesirable to operate at an earlier age.

We believe there is an important role for laypersons' subjective impressions of appearance in research on cranioplastic outcomes for children with SSC. For example, appearance ratings could be used to establish the social validity of various cranial index quantifications of SCC taken from radiographic images,^{19,20} allowing for greater precision in determining what are "acceptable" surgery outcomes. Ultimately, the combination of quantitative measures of imaging data, clinician ratings, and ratings by naïve observers may all be used to compare the relative effects of different surgical procedures.

Limitations of this study include the small number of participants with different SSC diagnoses, precluding a more detailed evaluation of differences as a function of suture fused. Photographs were missing or of poor quality for a relatively high proportion of study participants. However, it does not appear that findings were biased by missing data, and results were unchanged when we restricted analyses to the sub-sample of participants with photographs at all three time points. We limited this study to ratings of front-view facial

photographs, which may not fully capture the social perception of appearance in person. If anything, this would be expected to underestimate group differences, as anomalies in head shape are likely to be more apparent from side and top views. Surgical data were not collected for all participants, making it impossible to compare outcomes as a function of the specific surgical procedure completed. Finally, although case-control differences were consistent and large in magnitude, this study did not examine the social or clinical significance of these differences. In addition to knowing whether a child looks different than his or her peers, it is important to know whether they are likely to be teased or stigmatized due to those differences. We will be able to examine those questions when children from our sample reach school-age.

CONCLUSIONS

Our findings suggest that children with SSC receive lower appearance ratings than unaffected peers, and that these differences remain following surgery to improve overall head shape. After adjusting for site of suture fusion, better outcomes in appearance were associated with earlier age at surgery. Further research is needed to determine whether other surgical variables (e.g., type of surgical procedure) might predict outcomes in appearance. We are also conducting an age 7 follow-up of the participants from this cohort, which will allow us to examine longer-term cosmetic and morphological outcomes and to determine the clinical significance of differences in appearance.

Acknowledgments

Sources of Funding:

This work was supported by a grant from the National Institute of Dental and Craniofacial Research (NIDCR grant # R01 DE 13813 awarded to Dr. Speltz).

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Figure 1. Sample photographs, used for appearance ratings, of a participant with sagittal synostosis from infancy through age 36 months.

Table 1

Baseline demographic characteristics of subjects with and without single-suture craniosynostosis with at least one measure of average facial attractiveness

Demographic Characteristic	Controls		Total Cases	
	Number	Percent	Number	Percent
Total	186	100.0%	193	100.0%
Sex				
Female	73	39.2%	70	36.3%
Male	113	60.8%	123	63.7%
Age group (months)				
< 6	83	44.6%	89	46.1%
6–9	56	30.1%	51	26.4%
9	47	25.3%	53	27.5%
Race				
Non-white or Hispanic	51	27.4%	54	28.0%
White, Non-Hispanic	135	72.6%	139	72.0%
SES Category				
I (highest)	55	29.6%	47	24.4%
II	93	50.0%	75	38.9%
III	26	14.0%	39	20.2%
IV	10	5.4%	24	12.4%
V (lowest)	2	1.1%	8	4.1%
Site				
Seattle	84	45.2%	81	41.3%
Chicago	58	31.2%	69	35.2%
St. Louis	8	4.3%	7	3.6%
Atlanta	34	18.3%	32	16.3%
Philadelphia	2	1.1%	7	3.6%
Affected suture				
Sagittal	-	-	83	43.0%
Metopic	-	-	51	26.4%
Unicoronal	-	-	49	25.4%
Lambdoid	-	-	10	5.2%
Age at surgery (months)				
1–6	-	-	73	37.8%
7–12	-	-	91	47.2%
13–18	-	-	17	8.8%
19+	-	-	12	6.2%

* Age at surgery missing for 3 participants

Table 2

Distribution of age at surgery among subjects with single-suture craniosynostosis, by affected suture

Suture site	n	mean	sd	minimum	maximum
<i>Sagittal</i>	83	7.07	6.12	1.81	34.13
<i>Metopic</i>	51	9.64	4.04	3.95	25.72
<i>Unicoronal</i>	49	11.93	5.34	4.74	28.21
<i>Lambdoid</i>	10	9.69	1.63	7.10	11.64

* Age at surgery missing for 3 participants

Table 3

Average appearance ratings for children with and without single-suture craniosynostosis by affected suture at Time1, Time 2, and Time 3.

	Time 1			Time 2			Time 3		
	n	Mean	sd	n	Mean	sd	n	Mean	sd
Controls	104	4.84	0.67	104	4.95	0.69	157	4.55	0.65
All Cases	158	4.14	0.72	157	4.39	0.75	132	3.97	0.77
<i>Sagittal</i>	71	4.23	0.72	71	4.64	0.68	55	4.16	0.75
<i>Metopic</i>	44	4.18	0.63	44	4.26	0.72	39	3.87	0.73
<i>Unicoronal</i>	36	3.93	0.73	36	4.09	0.69	32	3.80	0.83
<i>Lambdoid</i>	7	3.95	1.03	6	4.21	1.24	7	3.79	0.64

Table 4

Differences in average appearance ratings between subjects with and without single-suture craniosynostosis over time

	# Cases/Controls	Adjusted Diff ^a	Group Differences 95% CI (Upper, Lower)	p-value
Time 1	158 104	-0.70	-0.87 -0.53	<0.001
Time 2	157 103	-0.55	-0.72 -0.37	<0.001
Time 3	132 157	-0.58	-0.75 -0.42	<0.001
Case-Control Differences in Trajectories				0.16

^a Adjusted for age (months), sex, study site, and race/ethnicity