OPEN

PRS GLOBAL OPEN

ORIGINAL ARTICLE
Craniofacial/Pediatric

Evaluating Surgical Decision-making in Nonsyndromic Sagittal Craniosynostosis Using a Digital 3D Model

Christopher D. Hughes, MD, MPH*
Olivia Langa, BA†
Laura Nuzzi, BA†
Steven J. Staffa, MS‡
Mark Proctor, MD†
John G. Meara, MD, DMD, MBA†
Ingrid M. Ganske, MD, MPA†

Background: Surgical correction of craniosynostosis addresses potentially elevated intracranial pressure and the cranial deformity. In nonsyndromic sagittal synostosis, approximately 15% of patients have elevated intracranial pressure. The decision to operate therefore likely reflects a combination of aesthetic goals, prevention of brain growth restriction over time, surgeon training and experience, and parental expectations. This study examines clinical factors that influence surgical decision-making in nonsyndromic sagittal synostosis.

Methods: An online survey sent to craniofacial and neurosurgeons presented 5 theoretical patients with varying severities of sagittal synostosis. For each cephalic index, 4 separate clinical scenarios were presented to assess influences of parental concern and developmental delay on the decision to operate.

Results: Fifty-six surveys were completed (response rate = 28%). Participants were predominantly from North America (57%), had over 10 years of experience (75%), and performed over 20 craniosynostosis procedures annually (50%). Thirty percent of respondents indicated they would operate regardless of head shape and without clinical and/or parental concern. Head shape was the greatest predictor of decision to operate ($P < 0.001$). Parental concern and developmental delay were independently associated with decision to operate ($P < 0.001$). Surgeons with more experience were also more likely to operate across all phenotypes (OR: 2.69, $P < 0.004$).

Conclusions: Surgeons responding to this survey were more strongly compelled to operate on children with nonsyndromic sagittal craniosynostosis when head shape was more severe. Additional factors, including parental concern and developmental delay, also influence the decision to operate, especially for moderate phenotypes. Geographic and subspecialty variations were not significant. *(Plast Reconstr Surg Glob Open 2021;9:e3493; doi: 10.1097/GOX.0000000000003493; Published online 21 May 2021.)*

INTRODUCTION

Sagittal craniosynostosis is the most common form of nonsyndromic craniosynostosis.[1] In unaffected patients without craniosynostosis, the morphology of the cranium is related to the underlying functional effects of the brain and dura: bones grow and head shape forms in the presence of patent cranial sutures.[2] With premature sutural fusion, cranial morphology takes on characteristic shapes corresponding to the particular fused suture(s). Children with sagittal craniosynostosis exhibit varying degrees of scaphocephaly, which may include frontal bossing, an anteriorly displaced vertex, narrowed bi-parietal distance, and a bullet-shaped occiput.[3] The effect on head shape is more significant when fusion occurs early in development.[2,4] The head circumference typically increases as a result of excessive anomalous anterior-posterior skull growth.[5]

In all cases of craniosynostosis, surgical treatment is indicated for elevated intracranial pressure (ICP) and/or substantial alterations in normal anatomy and appearance. The risk of elevated ICP in single suture, nonsyndromic sagittal synostosis is estimated to be 10%–20%,[6] but reports are inconclusive and often contradictory. Data regarding the probability of neurodevelopmental delay associated with the condition are likewise inconsistent; some suggest up to a 5-fold increased risk for developmental delays and

*From the *Division of Plastic and Craniofacial Surgery, Connecticut Children's, Hartford, Conn.; †Department of Plastic and Oral Surgery, Boston Children's Hospital, Boston, Mass.; and ‡Department of Anesthesiology, Critical Care and Pain Medicine, Boston Children's Hospital, Boston, Mass.*

Received for publication September 30, 2020; accepted January 26, 2021.

DOI: 10.1097/GOX.0000000000003493

Disclosure: *The authors have no financial interest to declare in relation to the content of this article.*

learning deficits in infancy.[7–9] Although there are numerous approaches in treating isolated sagittal craniosynostosis, there is currently no consensus among craniofacial surgeons or pediatric neurosurgeons regarding the optimal approach.[10,11] Furthermore, the decision whether to operate at all in nonsyndromic patients with sagittal synostosis relies largely on intuition and individual surgical judgment, without objective data to clarify clinical management. Others have investigated similar decision-making processes for other forms of craniosynostosis.[12]

The goal of this study was to identify factors that prompt in cases of sagittal synostosis surgical intervention. Using a custom-engineered 3D design framework, we developed a novel survey tool to assess practice patterns among pediatric neurosurgeons and pediatric craniofacial surgeons around the world.

METHODS

Scenario Descriptions

Five hypothetical 11-month-old patients with varying cephalic index (CI) severities were created: 81 (normal), 78 (minor), 71 (moderate), 68 (severe), and 65 (very severe). Eleven months was selected as a representative age by which a child would have been referred for evaluation and at which it would be reasonable to proceed with calvarial remodeling if a surgeon was inclined to offer this.

For each phenotypic severity, 4 scenarios were created. The first scenario was considered baseline: a child without delay in developmental milestones and with parents who were not explicitly concerned about the head shape. Patient scenario 2 included the addition of parental concern about head shape, with all other factors (cephalic index, growth chart, CT scans, and developmental delay) unchanged. Patient scenario 3 consisted of the child "not yet sitting independently," indicating a delay in motor development, with all other factors unchanged. The final scenario, scenario 4, included findings from both scenario 2 and 3—parents were concerned about the head shape, and there was gross motor delay.

Virtual Patient Creation

Digital models for each of the 5 hypothetical patients were created using Maya software (version 2018.5, 2019; Autodesk Inc. San Rafael, Calif.) (Fig. 1). These were designed based on a typical patient presentation for each cephalic index severity. The renderings, rather than actual patient photographs, were used to reduce distractors (ie, hair, lighting) and potential confounders (ie, facial features or expressions, ethnic or racial background). Models

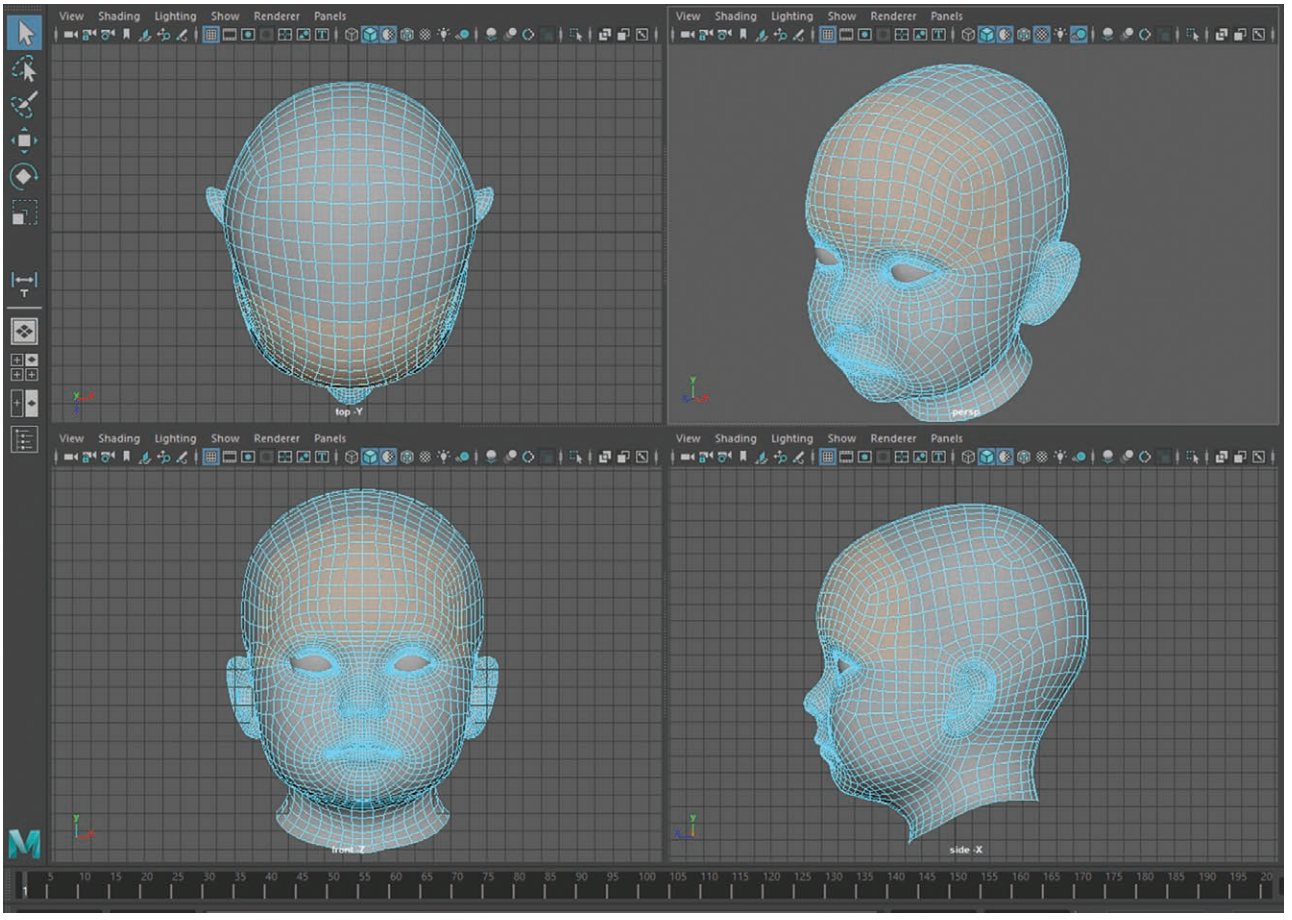

Fig. 1. Phenotype model production. Representative example of design matrix used in the creation of the customized, 3D framework for evaluating progressive scenarios of sagittal craniosynostosis.

were refined based on iterative feedback provided by the craniofacial surgeons at our institution.

Survey Design

Each set of patient images was paired with a corresponding growth chart, 3D reconstructions of CT scans taken from actual patients, and details of the hypothetical patient's age and sex, developmental status, whether parents were concerned about the head shape, and the cephalic index. A clinical presentation slide was created for each of the 4 scenarios, for each of the 5 patients, totaling 20 slides (Fig. 2).

This novel survey was built using a Research Electronic Data Capture (REDCap) database (REDCap 8.10.18; Vanderbilt University).[13] Various survey iterations were performed after internal review was done among our Craniofacial team, including input from Plastic Surgery and Pediatric Neurosurgery. Hypothetical patients were presented in an arbitrary order to avoid potential predictive bias when presented with a progressively severe patient phenotype. For each patient presentation slide, the participant was asked "how would you surgically manage this child?" with options to operate or observe. If the participant chose operate, options of typical craniosynostosis procedures were offered. If the participant selected to observe, the participant was asked if they would order any adjunctive tests: ophthalmologic exam, MR, or ICP monitoring. If the participant selected additional tests, the results were found to be unremarkable, confirming a lack of elevated ICP. The participants were again asked whether they would choose operate based on the clinical information or continue observation.

Survey branching logic was utilized to minimize redundancy. Participants who selected operation on the very first scenario of a patient (ie, based on baseline head shape and CI alone, without additional concern from the parents and without developmental delay) would presumably offer an operation for the same CI with the addition of the other clinical factors. Thus, if a participant initially selected operation, they were then shown the next patient severity, skipping the other scenarios for that specific patient example.

Participants

Participants were identified through the International Society for Craniofacial Surgery database and using personal and public records of neurosurgeons and craniofacial surgeons. The survey was delivered to 198

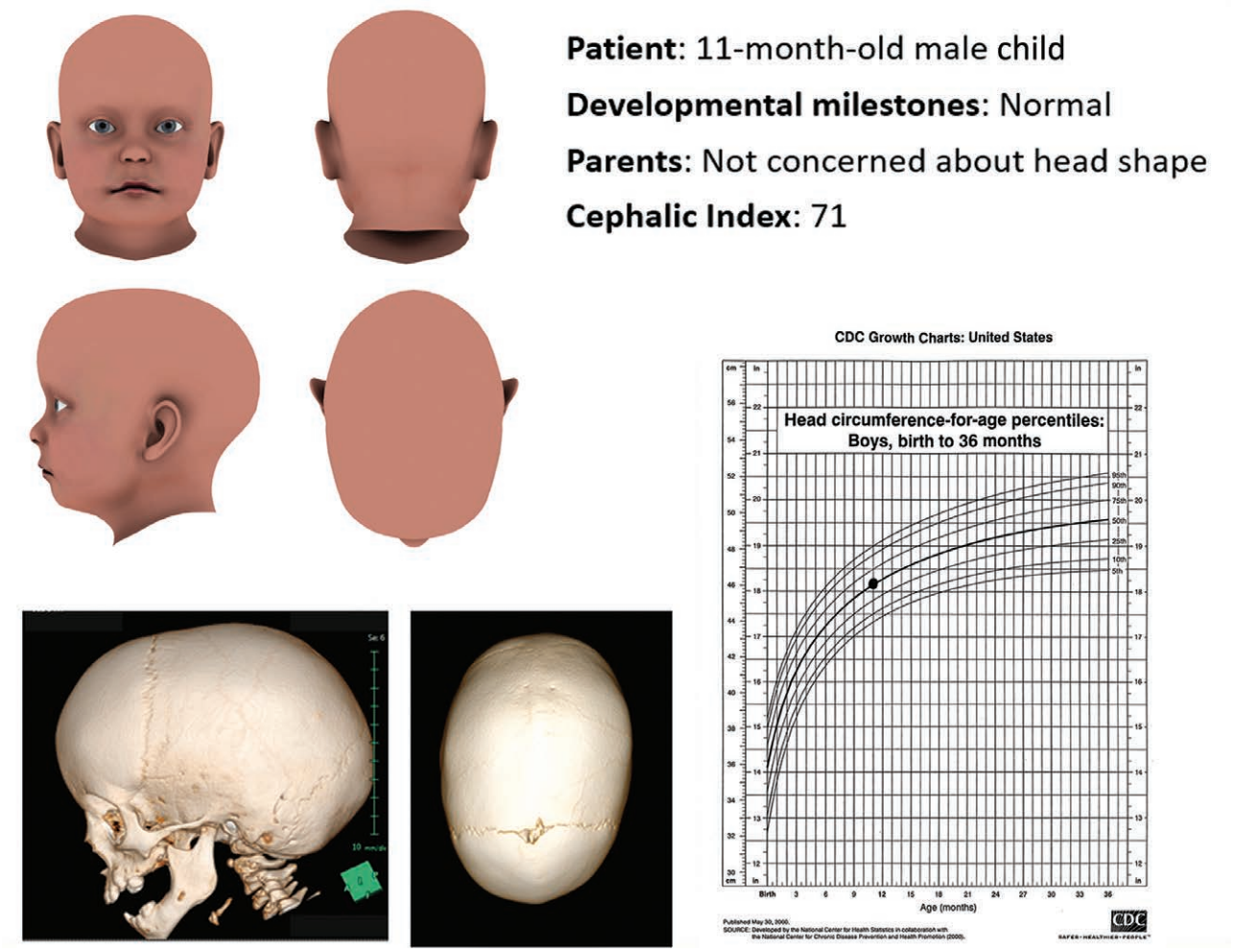

Fig. 2. Survey patient example. Representative example of a patient scenario, as presented in the survey.

participants: 86% were craniofacial surgeons and 9% were neurosurgeons. Participants were recruited via email message, including a link to the online survey. Survey reminders were sent 4 and 7 weeks later. All participants were de-identified. Demographic information was collected regarding surgical specialty, years in practice, number of craniosynostosis procedures performed each year, and geographic region of practice.

Statistical Analysis

Categorical data on demographics and rates of decision to operate within respondent subgroups are presented as frequencies and percentages. For each case scenario type, the Cochran-Armitage test was used to evaluate linear trends in operation rates across the cephalic index types. A multivariable mixed-effects logistic regression analysis was performed via generalized estimating equations to evaluate the independent associations between cephalic index, scenario, and surgeon factors and the decision to operate. Surgeon respondent ID was incorporated as a random effect into the modeling to account for the correlation between multiple responses from the same surgeon, whereas all other variables were included as fixed effects. The results from the mixed-effects model are presented as adjusted odds ratios, with corresponding 95% confidence intervals and *P* values. Statistical analyses were performed using SPSS Statistics for Windows version 24.0 software (IBM Corp, Armonk, N.Y.) and Stata version 16.0 (StataCorp LLC, College Station, Tex.). A two-tailed $P < 0.05$ was used to determine statistical significance.

RESULTS

Participant Demographics

Fifty-six surveys (56/198; 28% response rate) were fully completed within the study period. Eighty-four percent of participants (n = 47) were craniofacial surgeons and 16% (n = 9) were neurosurgeons. The majority of surgeons had been in practice for over 10 years (n = 42, 75%). The remainder included 12.5% (n = 7) each, who had been in practice for 6–10 years or < 5 years. Half (50%) of the respondents performed 20 or more craniosynostosis operations per year, and <10% of respondents did fewer than 5 per year. Fifty-seven percent of respondents were from North America (n = 32), with fewer from Europe (n = 11; 19%), Asia (n = 6; 11%), and South America (n = 6; 11%) (Table 1).

Table 1. Demographics of Survey Participants (n = 56)

Demographics	
Specialty	
Craniofacial	47 (84%)
Neurosurgery	9 (16%)
Years of practice	
<5	7 (12.5%)
6–10	7 (12.5%)
>10	42 (75%)
Number of operations per year	
<5	5 (9%)
6–10	8 (14%)
11–20	15 (27%)
>20	28 (50%)
Region	
Africa	0
Asia	6 (11%)
Europe	11 (19%)
Middle East	0
North America	32 (57%)
South America	6 (11%)
Other	1 (2%)

Severity of Cephalic Index

Thirty percent (n = 17) of the survey participants indicated that they would operate on the virtual patient with sagittal synostosis regardless of a normal appearing head shape and lack of clinical and/or parental concerns (Table 2). With each incremental decline of the cephalic index, an increasing number of respondents indicated that they would operate. At the most severe phenotype (CI 65), 96% (n = 54) would operate, even in the absence of parental and/or developmental concerns. In our multivariable regression analysis, the cephalic index was independently associated with the odds of selecting to operate (Table 3). Compared with baseline CI 81, the adjusted odds for the decision to operate were significantly higher for CI 78 (OR = 1.41; 95% CI: 1.02, 1.95; $P = 0.04$), for CI 71 (OR = 5.6; 95% CI: 3.86, 8.13; $P < 0.001$), for CI 68 (OR = 14.9; 95% CI: 9.18, 24; $P < 0.001$), and for the most severe phenotype (CI 65) (OR = 125.9; 95% CI 38.3, 415; $P < 0.001$).

Parental Concern and Developmental Delay

The presence of parental concern and/or developmental delay significantly increased the likelihood that the respondent would elect to operate when compared with the baseline scenario (Table 3). When the patient presented with parental concern about the head shape, participants were 2.29 times more likely to choose to operate than for the baseline scenario (OR = 2.29; 95% CI: 1.59, 3.30; $P < 0.001$). When developmental delay was present, participants were 1.65 times more likely to operate compared with the baseline scenario (OR = 1.65; 95% CI: 1.15, 2.35; $P = 0.006$). Although not significantly different, participants had 1.39 times the odds of choosing to operate when parents were concerned compared with the developmental delay scenario (OR = 1.39; 95% CI: 0.96, 2.01; $P = 0.08$). When parental concern and developmental delay were both present, participants were 3 times more likely to choose to operate than at the baseline (OR = 3; 95% CI: 2.06, 24.37; $P < 0.006$). There was a statistically significant linear trend in which respondents' decision to operate increased as cephalic index and head shape became more severe, for each scenario ($P < 0.001$, all; Fig. 3).

Surgeon Experience

Surgeons practicing >10 years were associated with 2.69 times the likelihood to operate overall compared with surgeons with fewer years of practice (OR = 2.69; 95% CI: 1.37, 5.29; $P = 0.004$) (Table 3). Compared with surgeons with less experience, a greater proportion of more experienced surgeons chose to operate on children in the minor phenotypes (CI: 81 and 78). All surgeons demonstrated significant linear trends in which their decision to

Table 2. Decision to Operate by Phenotype and Scenario

Cephalic Index	81	78	71	68	65
Baseline					
Operation	17 (30%)	18 (32%)	39 (70%)	48 (86%)	54 (96%)
Observation	39 (70%)	38 (68%)	17 (30%)	8 (14%)	2 (4%)
Parents concerned					
Operation	24 (43%)	31 (55%)	47 (84%)	52 (93%)	56 (100%)
Observation	32 (57%)	25 (45%)	9 (16%)	4 (7%)	0 (0%)
Developmental delay					
Operation	26 (46%)	28 (50%)	40 (71%)	48 (86%)	55 (98%)
Observation	30 (54%)	28 (50%)	16 (29%)	8 (14%)	1 (2%)
Parents concerned and developmental delay					
Operation	28 (50%)	35 (63%)	48 (86%)	53 (95%)	56 (100%)
Observation	28 (50%)	21 (38%)	8 (14%)	3 (5%)	0 (0%)

Table 3. Multivariable Mixed Effects Logistic Regression Analysis of Decision to Operate

Covariate	Adjusted Odds Ratio	95% Confidence Interval	*P*
Cephalic index			
81	Reference	—	—
78	1.41	(1.02, 1.95)	0.04*
71	5.60	(3.86, 8.13)	<0.001*
68	14.90	(9.18, 24.00)	<0.001*
65	125.90	(38.30,414.60)	<0.001*
Scenario			
Baseline	Reference	—	—
Parents concerned	2.29	(1.59, 3.30)	<0.001*
Developmental delay	1.65	(1.15, 2.35)	0.006*
Parents concerned and developmental delay	3.00	(2.06, 4.37)	<0.001*
Year in practice			
≤10	Reference	—	—
>10	2.69	(1.37, 5.29)	0.004*
Region of practice			
Asia	1.59	(0.65, 3.89)	0.304
Europe	0.53	(0.25, 1.13)	0.101
North America	Reference	—	—
South America	1.95	(0.79, 4.81)	0.146
Specialty			
Craniofacial surgery	Reference	—	—
Neurosurgery	1.03	(0.47, 2.24)	0.947

Adjusted odds ratios, 95% confidence intervals, and P values for the likelihood of operating were calculated using generalized estimating equations modeling to account for multiple responses for each cephalic index and scenario by the same surgeon respondent.
*Statistically significant.

operate increased as the CI increased in severity under each scenario ($P < 0.001$) (Fig. 4). Surgical subspecialty was not independently associated with the decision to operate ($P = 0.95$).

Geographic Trends

Respondents were primarily from North America and Europe (Table 1). Geographic location was not a significant predictor of respondents' decision to operate. European surgeons were not more or less likely to operate than their North American counterparts (OR = 0.53; 95% CI: 0.25, 1.13; $P = 0.10$) (Table 3).

DISCUSSION

The goal of this study was to examine specific factors that influence the decision to operate on patients with nonsyndromic sagittal craniosynostosis. We surveyed craniofacial surgeons and pediatric neurosurgeons worldwide using a customized, 3D simulation tool that provided a spectrum of phenotypic and clinical characteristics for a hypothetical patient with sagittal craniosynostosis.

Overall, we confirmed a lack of standardized approach to care for patients with sagittal synostosis. This study found that head shape, quantified by cephalic index, was the most significantly predictive factor in deciding to operate for all survey participants. Additional clinical variables such as parental preferences and motor delays contributed to operative decision-making, especially in the more minor-to-moderate phenotypes. There was some influence of surgeon experience on operative decision-making; surgeons with less experience were less likely to operate on minor phenotypes. There was no influence of geographic region or surgical specialty on the decision to operate.

Severity of Cephalic Index

Across all groups, severity of scaphocephaly (increasing CI) was found to be the most predictive factor in surgeons' decision to operate on nonsyndromic sagittal synostosis. Regardless of parental concerns or additional clinical factors, the decision to operate was more common as the head shape lengthened. CI is the most commonly used outcome measure to evaluate success following cranioplasty for sagittal synostosis.[14] There are data to suggest regional and ethnic variations in CI,[15,16] and studies have documented that average cephalic indices have changed with introduction of the Back to Sleep campaign.[17]

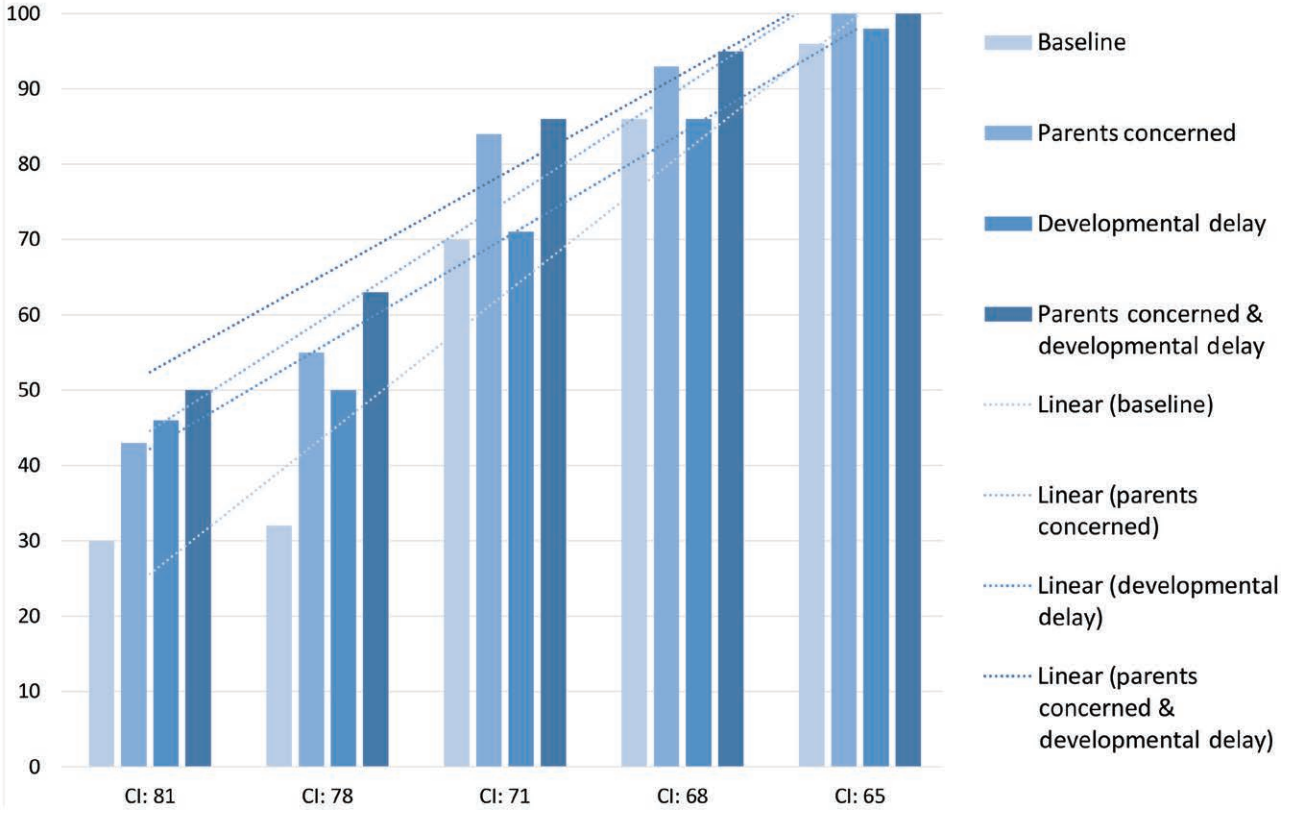

Fig. 3. Linear relationships between decisions to operate for each clinical scenario. Each color bar represents a unique clinical scenario for a given cephalic index (x-axis). Across all cephalic indices, the trends toward operation (dotted lines) were statistically significant (Cochran-Armitage test, $P < 0.001$ for each scenario).

Despite being commonly used as an indicator of phenotype, CI alone may not be an ideal proxy for severity. In an elegant analysis of the utility of CI, Fearon and colleagues evaluated the preoperative CI measurements for patients with isolated sagittal craniosynostosis, as well as the values for patients who had previously undergone an operation who later presented with abnormal head shape requiring a secondary operation. Overall, his group found that CI was not significantly abnormal in the preoperative group and that it was not a predictive measure of the need for revisional procedure. A third component of that study evaluated CI in a photography-based assessment of head shape and found no correlation between appearance and cephalic index.[18] Others have suggested alternative measures of CI that may more accurately account for euryon displacement seen in craniosynostosis.[19–21] For this reason, we used the series of model images as well as CI measurements to characterize the phenotypes in this study. Given the nature of our survey design, we are unable to determine whether respondents made decisions based on the appearance of the child's head itself or on the corresponding CI attributed to it, or both.

Other groups have investigated surgeons' preferences with respect to management for patients with sagittal craniosynostosis.[10,11] These survey-based investigations were largely focused on procedural and technical details in management, but 1 group did note that 63% of respondents self-reported that "skull deformity" was the primary indication for the surgical treatment of sagittal craniosynostosis.[11] Data regarding the metrics by which this assessment was made were not included in their analysis. Our data corroborate the fact that appearance of the skull is the most substantial factor in surgical decision-making.

Parental Influence

Our study also investigated the role that parental preference may have in the decision to operate on children with nonsyndromic sagittal synostosis. In our analysis, parental concern was a significant independent predictor of a surgeon's decision to operate, and it seemed to have its greatest effect when combined with evidence of developmental delay. We did see a trend in the data at more minor phenotypes: a greater proportion of surgeons chose to operate when parents were concerned and when there was evidence of developmental delay at the more minor phenotypic range. These data suggest that when the scaphocephaly is not severe, parental preferences play a larger role in the decision to operate.

There are limited data within the literature evaluating parental preferences with respect to sagittal craniosynostosis. A survey of the general population (not specifically parents of children with sagittal craniosynostosis) demonstrated that people would be willing to trade an 8% risk of death or 3 years of life to live without scaphocephaly.[22]

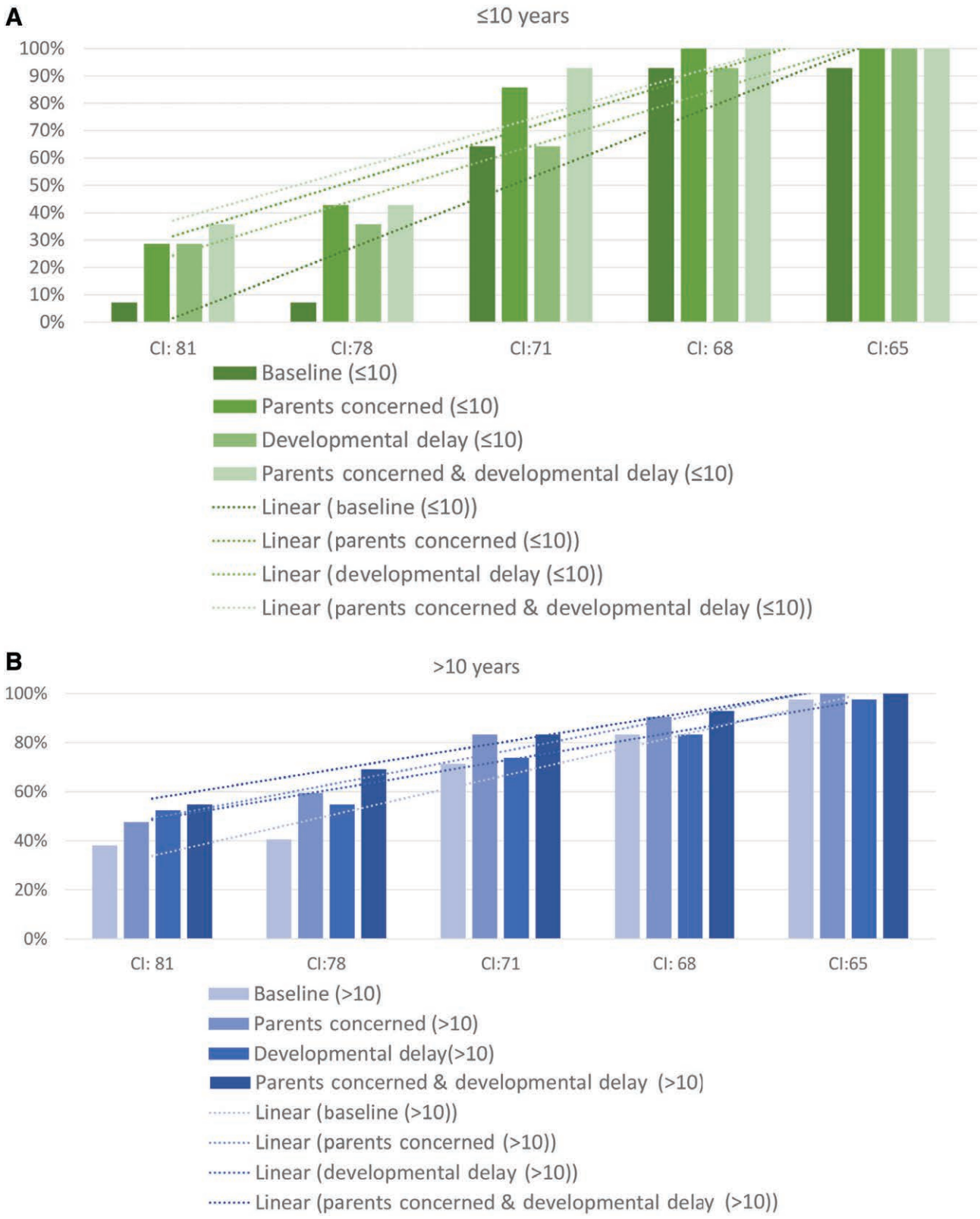

Fig. 4. Surgeon experience. A, Results for surgeons with 10 or fewer years of experience. B, Results for surgeons with >10 years of experience. Each color bar represents a unique clinical scenario for a given cephalic index. Across all cephalic indices, the trends toward operation (dotted lines) were statistically significant (Cochran-Armitage test, $P < 0.001$ for each scenario).

These data represent personal choices and do not accurately reflect parental decision-making, but the findings are useful in highlighting the discordance between perception of the diagnosis and the natural history of the condition.[23] This discrepancy seems to be present in surgeons as well; interestingly, almost one third of our respondents reported that they would operate on a child with sagittal synostosis and a normal head shape and CI. Our current data suggest that parental preferences can significantly contribute to surgical decision-making around craniosynostosis. Further objective outcome studies on the long-term outcomes and natural history of the condition may improve shared surgical decision-making with parents.

Developmental Delay

Enhancing parental education is especially important given the lack of clarity regarding intellectual and developmental delays among patients with sagittal synostosis.[24] Intracranial volume (ICV) is often used as a surrogate for potential elevation in intracranial pressure; however, there is a lack of consensus on whether Intracranial volume decreases with sagittal craniosynostosis, with some studies suggesting it does[25,26] and others finding it does not.[27–29] Likewise, data regarding neurodevelopmental delay for patients with isolated sagittal craniosynostosis are often contradictory as well. Several studies suggest a notable developmental delay for children with isolated sagittal craniosynostosis, especially when compared with unaffected children.[1,9,30,31] In a longitudinal, multicenter study comprising data from 5 craniofacial centers across the USA, Starr and colleagues found that children with treated sagittal craniosynostosis had 1.5–2× the odds of being delayed on the cognitive, motor, and speech scales.[30] Other studies, however, suggest that children with isolated sagittal craniosynostosis do not demonstrate inferior intellectual outcomes at older ages.[32]

We introduced the neurodevelopmental delay concept within our model as an isolated motor delay in our 11-month-old theoretical patient. Developmental delay was a significant independent predictor of a surgeon's decision to operate but was not as impactful as parental concern. The combination of both parental concern and developmental delay significantly contributed to the surgeon's decision to operate. These results likely reflect variations in the interpretation of the data regarding neurodevelopmental delays for patients with isolated sagittal craniosynostosis among our craniofacial and pediatric neurosurgical respondents.

Surgeon Experience and Geographic Trends

We found that surgeons with more than 10 years of experience were more likely to operate overall and on patients with less severe scaphocephaly. There is a relative dearth of data regarding surgeon experience and operative decision-making in the craniofacial literature, but a recent survey of orthopedic surgeons suggests that more experienced surgeons tended to operate less frequently when the indications for operation were equivocal.[33] Our data differ from those findings and suggest that surgeons with less experience may be less willing to undertake a large operation with its attendant risks for a more minor expected postoperative improvement, or perhaps are trained under differing paradigms than their more senior colleagues. Conversely, surgeons with more experience may be more comfortable operating on children with more minor phenotypic differences.

Limitations

This survey study is limited by the biases inherent in its design. We sampled a diverse group of surgeons who treat craniosynostosis, but the low response rate introduces potential for non-response bias in our findings. It is also possible that our responses are not wholly representative of the larger community of craniofacial surgeons and pediatric neurosurgeons around the world. The scenarios posed only a limited slate of factors that may play into surgical decision-making. Furthermore, when faced with the actual decision to operate rather than a hypothetical scenario, surgeons may ultimately make different decisions than what they indicate in a hypothetical scenario. In an effort to streamline the process, we surveyed a discrete number of clinical scenarios. Within our model design, there was also a limit to the granularity with which we could analyze our data. For instance, we were unable to determine if there were specific components of the child's head shape (bossing, low occiput) that were more influential than others in the decision to operate. The response rate from other regions outside of North America and Europe were underpowered to draw statistically meaningful conclusions regarding international variability.

CONCLUSIONS

Surgeons responding to this survey were more strongly compelled to operate on children with nonsyndromic sagittal craniosynostosis when the head shape was more severe. Other factors, including parental preferences and evidence of developmental delay, can influence operative decision-making. Neither surgical specialty nor geographic region is significantly associated with operative decision-making for sagittal craniosynostosis.

Ingrid M. Ganske, MD, MPA
Harvard Medical School
Department of Plastic and Oral Surgery
Boston Children's Hospital
300 Longwood Avenue
Boston, MA 02115
E-mail: ingrid.ganske@childrens.harvard.edu

ACKNOWLEDGMENT

The authors recognize Danger Donaghey for model production and artistic renderings. This study was approved by our institutional review board (IRB: P00030976).

REFERENCES

1. Kapp-Simon KA, Speltz ML, Cunningham ML, et al. Neurodevelopment of children with single suture craniosynostosis: a review. *Childs Nerv Syst.* 2007;23:269–281.

2. Sperber GH, Sperber SM, eds. *Craniofacial Embryogenetics and Development. Raleigh.* USA: PMPHUSA; 2018.
3. Hughes CD, Isaac KV, Hwang PF, et al. Modification of the Melbourne method for total calvarial vault remodeling. *Plast Reconstr Surg Glob Open.* 2018;6:e1848.
4. Mathijssen IM, van Splunder J, Vermeij-Keers C, et al. Tracing craniosynostosis to its developmental stage through bone center displacement. *J Craniofac Genet Dev Biol.* 1999;19:57–63.
5. Fearon JA. Evidence-based medicine: craniosynostosis. *Plast Reconstr Surg.* 2014;133:1261–1275.
6. Holland J, Rodrigues D, Mohan S, et al. Intracranial volume (ICV) in isolated sagittal craniosynostosis: a retrospective case-matched-control study. *Childs Nerv Syst.* 2019;35:509–516.
7. Knight SJ, Anderson VA, Spencer-Smith MM, et al. Neurodevelopmental outcomes in infants and children with single-suture craniosynostosis: a systematic review. *Dev Neuropsychol.* 2014;39:159–186.
8. Speltz ML, Kapp-Simon KA, Cunningham M, et al. Single-suture craniosynostosis: a review of neurobehavioral research and theory. *J Pediatr Psychol.* 2004;29:651–668.
9. Imahiyerobo TA, Johns AL, Christian EA, et al. Risk factors for preoperative developmental delay in patients with nonsyndromic sagittal craniosynostosis. *Plast Reconstr Surg.* 2019;143:133e–139e.
10. Lee BS, Hwang LS, Doumit GD, et al. Management options of non-syndromic sagittal craniosynostosis. *J Clin Neurosci.* 2017;39:28–34.
11. Doumit GD, Papay FA, Moores N, et al. Management of sagittal synostosis: a solution to equipoise. *J Craniofac Surg.* 2014;25:1260–1265.
12. Anolik RA, Allori AC, Pourtaheri N, et al. Objective assessment of the interfrontal angle for severity grading and operative decision-making in metopic synostosis. *Plast Reconstr Surg.* 2016;137:1548–1555.
13. Harris PA, Taylor R, Thielke R, et al. Research electronic data capture (REDCap)–a metadata-driven methodology and workflow process for providing translational research informatics support. *J Biomed Inform.* 2009;42:377–381.
14. Gerety PA, Basta MN, Fischer JP, et al. Operative management of nonsyndromic sagittal synostosis: a head-to-head meta-analysis of outcomes comparing 3 techniques. *J Craniofac Surg.* 2015;26:1251–1257.
15. Bharati S, Som S, Bharati P, et al. Climate and head form in India. *Am J Hum Biol.* 2001;13:626–634.
16. Bharati S, Demarchi DA, Mukherji D, et al. Spatial patterns of anthropometric variation in India with reference to geographic, climatic, ethnic and linguistic backgrounds. *Ann Hum Biol.* 2005;32:407–444.
17. Roby BB, Ferrello M, Huang TC, et al. Symptom timeline preceding cochlear implant failure: an institutional experience. *Otolaryngol Head Neck Surg.* 2012;146:782–787.
18. Fearon JA, Ditthakasem K, Herbert M, et al. An appraisal of the Cephalic index in sagittal craniosynostosis, and the unseen third dimension. *Plast Reconstr Surg.* 2017;140:138–145.
19. Dvoracek LA, Skolnick GB, Nguyen DC, et al. Comparison of traditional versus normative cephalic index in patients with sagittal synostosis: measure of scaphocephaly and postoperative outcome. *Plast Reconstr Surg.* 2015;136:541–548.
20. Ruiz-Correa S, Sze RW, Starr JR, et al. New scaphocephaly severity indices of sagittal craniosynostosis: a comparative study with cranial index quantifications. *Cleft Palate Craniofac J.* 2006;43:211–221.
21. Heller JB, Heller MM, Knoll B, et al. Intracranial volume and cephalic index outcomes for total calvarial reconstruction among nonsyndromic sagittal synostosis patients. *Plast Reconstr Surg.* 2008;121:187–195.
22. Kuta V, McNeely PD, Walling S, et al. Sagittal craniosynostosis: a utility outcomes study. *J Neurosurg Pediatr.* 2017;20:113–118.
23. Duhaime AC. Editorial. Sagittal craniosynostosis: what matters to parents? *J Neurosurg Pediatr.* 2017;20:111–112.
24. Chummun S, McLean NR, Flapper WJ, et al. The management of nonsyndromic, isolated sagittal synostosis. *J Craniofac Surg.* 2016;27:299–304.
25. Lee SS, Duncan CC, Knoll BI, et al. Intracranial compartment volume changes in sagittal craniosynostosis patients: influence of comprehensive cranioplasty. *Plast Reconstr Surg.* 2010;126:187–196.
26. Seeberger R, Hoffmann J, Freudlsperger C, et al. Intracranial volume (ICV) in isolated sagittal craniosynostosis measured by 3D photocephalometry: a new perspective on a controversial issue. *J Craniomaxillofac Surg.* 2016;44:626–631.
27. Fischer S, Maltese G, Tarnow P, et al. Intracranial volume is normal in infants with sagittal synostosis. *J Plast Surg Hand Surg.* 2015;49:62–64.
28. Toma R, Greensmith AL, Meara JG, et al. Quantitative morphometric outcomes following the Melbourne method of total vault remodeling for scaphocephaly. *J Craniofac Surg.* 2010;21:637–643.
29. Kolar JC, Salter EM, Weinberg SM. Preoperative craniofacial dysmorphology in isolated sagittal synostosis: a comprehensive anthropometric evaluation. *J Craniofac Surg.* 2010;21:1404–1410.
30. Starr JR, Collett BR, Gaither R, et al. Multicenter study of neurodevelopment in 3-year-old children with and without single-suture craniosynostosis. *Arch Pediatr Adolesc Med.* 2012;166:536–542.
31. Kapp-Simon KA, Wallace E, Collett BR, et al. Language, learning, and memory in children with and without single-suture craniosynostosis. *J Neurosurg Pediatr.* 2016;17:578–588.
32. Da Costa AC, Walters I, Savarirayan R, et al. Intellectual outcomes in children and adolescents with syndromic and nonsyndromic craniosynostosis. *Plast Reconstr Surg.* 2006;118:175–181.
33. Meunier A, Posadzy K, Tinghög G, et al. Risk preferences and attitudes to surgery in decision making. *Acta Orthop.* 2017;88:466–471.