Published in final edited form as:

J Craniofac Surg. 2014 July ; 25(4): 1226–1229. doi:10.1097/SCS.0000000000000875.

POINT OF MAXIMUM WIDTH: A NEW MEASURE FOR ANTHROPOMETRIC OUTCOMES IN PATIENTS WITH SAGITTAL SYNOSTOSIS

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Abstract

The aesthetic success of sagittal synostosis reconstruction is measured by cephalic index (CI). This limited measure does not fully account for the abnormal head shape in sagittal synostosis. In this retrospective study, we investigate a new objective measure, point of maximum width (PMW) of the skull from a vertex view, to determine where the head is widest for children with sagittal synostosis as compared to normal controls. Preoperative CT scans of 27 children with sagittal synostosis and 14 postoperative CT scans at least 8 months after surgery were obtained. Normal CT scans were matched for age, gender, and race. Three-dimensional renderings were standardized for orientation. Average PMW in patients with sagittal synostosis was $53\% \pm 1\%$ compared to $57\% \pm 1\%$ in controls ($p < 0.001$). Average CI in patients with sagittal synostosis was $66.8\% \pm 0.8\%$ compared to $83.3\% \pm 1.0\%$ in controls ($p < 0.001$). The correlation between PMW and CI was weak in both controls ($R^2 = 0.002$, $p = 0.824$) and uncorrected cases ($R^2 = 0.083$, $p = 0.145$). After surgical correction, both CI and PMW significantly improved. Average PMW in patients after surgical release of sagittal synostosis was $58\% \pm 1\%$ compared to $58\% \pm 1\%$ in controls ($p = 0.986$). PMW is not a surrogate for CI but is a novel, valid measure of skull shape, which aids in quantifying the widest region of the skull. PMW is significantly more anterior in children with sagittal synostosis and exhibits a consistent posterior shift along the cranium after surgery, showing no difference compared to healthy children.

Keywords

Craniosynostosis; sagittal synostosis; point of maximum width

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Introduction

Isolated sagittal synostosis, the pathologic abnormal fusion of the sagittal suture, is the most common type of single-suture craniosynostosis.¹ Premature closure of the sagittal suture results in a classic head shape, including scaphocephaly (elongation of the skull in the antero-posterior dimension), variable degrees of frontal bossing, coronal constriction, occipital protuberance, and bitemporal protrusion.² Diagnosis relies on physical examination of the scaphocephalic head shape and is verified by plain skull radiographs or computed tomography (CT) scans of the head.

Surgical release of the sagittal suture is the mainstay of treatment for sagittal synostosis and can be accomplished by strip craniectomy, open calvarial vault remodeling (CVR), endoscopic suture release, or spring-mediated cranioplasty.^{3, 4} Surgical intervention potentially allows for normal brain growth and development, in addition to correcting the aesthetic skull deformity, thereby improving psychosocial development.⁴ The aesthetic success of surgical release of sagittal synostosis is currently measured by a single objective marker, cephalic index (CI), which is a ratio of width to length of the cranium expressed as a percentile. CI does not fully account for the abnormal head shape in children with sagittal synostosis, as it only addresses the dolichocephalic component of the skull.

The purpose of this study is to introduce and validate a new objective measure, point of maximum width (PMW) of the skull from a vertex view, to determine if the head is widest at different places for children with sagittal synostosis as compared to healthy children. In this study, we hope to show that PMW is a valid and reproducible metric obtained from CT scans, which contributes additional information to that provided by CI. Moreover, the authors hypothesize that surgical correction of sagittal synostosis normalizes PMW, as well as CI.

Material and Methods

Institutional review board approval was obtained prior to initiation of the study. The Washington University in Saint Louis School of Medicine Craniofacial Database was used to identify appropriate patients for this study. Patients who had a diagnosis of isolated sagittal synostosis and underwent surgical repair between 1987 and 2006 at Saint Louis Children's Hospital were evaluated in the initial query. Any patient with an additional craniofacial diagnosis that could potentially affect skull and facial development was excluded. Patients enrolled in this study had a preoperative CT scan and a subsequent postoperative CT scan between 8 and 14 months after surgery. If CT scan data was not available for both of these time points, or the identified CT scans had any significant motion artifact or inadequate detail to complete the analysis, those patients were excluded. The remaining patients were classified by type of surgical procedure. For the sake of uniformity, only patients that underwent open CVR were included. Patients treated with endoscopic-assisted release, strip craniectomy, or spring-release were excluded from this study.

Computed tomography scans were obtained from the Radiology Database at Saint Louis Children's Hospital to establish age-, gender-, and race-matched normal controls. These CT

scans showed no underlying traumatic or congenital craniofacial deformity. Patient age between cases and controls was matched within 2 weeks. This left 27 preoperative and 14 postoperative CT scans with a diagnosis of sagittal synostosis who had appropriately matched normal controls in the craniofacial database.

The CT scan data were translated into three-dimensional reconstructions using Analyze 11.0 (Mayo Clinic, Rochester, MN). Only bone windows were used when orienting the craniofacial skeleton. A few of the CT scans analyzed did not include the inferior orbital rim, making orientation to the Frankfort horizontal plane impossible in those cases. Instead, the sellanasion plane was used as the horizontal plane to standardize the orientation of all CT images (Figure 1). Images were then rotated to a vertex view to enable measurements of length and width.

Cephalic index (CI), the ratio of skull width to length expressed as a percentile was obtained for both sagittal synostosis patients and their normal controls. The length of the skull was defined as the maximal antero-posterior distance along the midsagittal plane and the width of the skull was calculated as the maximum biparietal diameter (euryon-euryon) perpendicular to this midsagittal plane (Figure 2). CI was calculated as the BP diameter divided by the AP diameter, multiplied by 100.

A novel measure, point of maximum width (PMW), was also calculated. PMW is defined as the ratio of the distance from the glabella to the maximum width of the skull in the midsagittal plane divided by the total length of the skull (Figure 3). Statistical analyses were performed using Microsoft Excel and SPSS v. 20 (IBM, Chicago, IL) and included Student's paired t-tests and Pearson's correlation calculations.

Results

The average age of the 27 patients with a CT scan at preoperative presentation for sagittal synostosis was 9.6 ± 3.2 months (mean $\pm$ standard error). Three of the 27 patients presented in a delayed fashion, over a year-old at initial presentation. The average preoperative age of patients excluding these three outliers was 4.6 ± 0.4 months. The average age of the 14 patients with at least 8 months of follow-up after surgical correction was 29.2 ± 6.6 months. For the preoperative group, 2/27 (7.3%) were African-American and 25/27 (92.6%) were Caucasian. For the postoperative group, the race distribution was similar: 1/14 (7.1%) African-American and 13/14 (92.9%) Caucasian. Gender distribution was essentially equivalent between the preoperative and postoperative groups: 6/27 (22.2%) versus 3/14 (21.4%) females, respectively.

The range of PMW amongst normal controls was 50.7% to 63.5%, with a mean of $57.2\% \pm 0.5\%$. Figure 4 shows the distribution of PMW for uncorrected cases of sagittal synostosis compared to normal controls obtained in children less than 12 months of age. Figure 5 shows the distribution of PMW for postoperative cases of sagittal synostosis compared to normal controls obtained from children between 14 and 92 months of age.

The average PMW in patients with sagittal synostosis was $53\% \pm 1\%$ compared to $57\% \pm 1\%$ in normal controls ($p < 0.001$). The average CI in patients with sagittal synostosis was

66.8% $\pm$ 0.8% compared to 83.3% $\pm$ 1.0% in normal controls ($p < 0.001$). Refer to Table 1. The correlation between PMW and CI was weak in both the controls ($R^2 = 0.002$, $p = 0.824$) and uncorrected cases ($R^2 = 0.083$, $p = 0.145$).

Approximately one year following surgical correction, both CI and PMW were significantly improved. The average PMW in patients after surgical release of sagittal synostosis was 58% $\pm$ 1%, compared to 58% $\pm$ 1% in normal controls ($p = 0.986$). PMW increased in 12 of 14 cases post repair. Figure 6 shows the change in PMW with surgical intervention. CI approached normal values post-repair ($p = 0.003$) with patients having a mean CI of 73.2% $\pm$ 1.1%. CI increased in all 14 cases. Refer to Table 2.

Discussion

Sagittal synostosis, the most common type of isolated sutural synostosis, occurs between 1 in 2000 to 5000 live births.¹ Surgical reconstruction is performed to improve the aesthetic appearance of the skull, relieve elevated intracranial pressure, and potentially allow for normal brain development.⁴ The best timing of surgical intervention and the most appropriate type of operation for correction are still being debated.^{3, 5}

Cephalic index (CI) by direct surface measurements is currently the most common method used to assess anthropometric outcomes after surgical reconstruction of sagittal craniosynostosis.⁶ It reflects the ratio of the cranial breadth as measured from widest point of the skull (euryon-euryon) to the cranial length as measured from glabella to opisthocranium (g-op), expressed as a percentile value.^{1, 7} Haas initially described the normative data based on plain radiographs, in an ethnically-mixed population, separated by age and gender. Normal values for cephalic index ranged from 76% to 83% for males and 76% to 84% for females.⁷ Due to the relatively longer antero-posterior dimension of scaphocephalic skulls, cephalic index is decreased to an average of 60-67% in sagittal synostosis.^{8, 9} Most centers, regardless of type of surgical intervention, report that cephalic index does increase and often normalizes for patients with isolated sagittal synostosis after surgical repair.¹⁰

While cephalic index does serve as an initial measurement of cranial shape, this metric alone is not sufficient to describe the abnormal skull shape seen in sagittal synostosis. It accurately assesses dolichocephaly (long, narrow head) but does not address other findings, such as the peaked appearance of the skull on the frontal view.¹¹ In some of our patients with “normal” cephalic indices postoperatively, skull shape remained irregular (Figure 7). Furthermore, patients who present with variable degrees of frontal bossing and occipital bossing will have a different maximal cranial length, as the glabella may no longer be the most anterior cranial point in these patients. With more asymmetric craniofacial findings, partial suture fusion, or patients with a delayed presentation, cephalic index is less accurate to describe cranial shape.

In this study, we analyzed an original metric (point of maximum width), as an additional marker for head shape, comparing children without craniofacial abnormalities to patients with an isolated diagnosis of sagittal synostosis before and after surgical repair. All patients

were age-, gender-, and race-matched to controls, and only patients who underwent open calvarial vault remodeling were studied. PMW is found to be significantly more anterior in children with sagittal synostosis prior to intervention compared to matched controls. While an exact normal value has yet to be determined, an estimation based on the normal CT scans included in this study places this value around 54-60%. While our sample size is relatively small (n = 41 controls), its gender distribution is similar to that seen in general sagittal synostosis population, with roughly a 4:1 ratio of males to females. The composition of our study population was predominantly Caucasian, so more ethnicities with larger sample sizes will need to be studied to confirm whether this measure is valid across races.

Following surgery, we noticed a consistent posterior shift in PMW along the cranium, with no significant difference in PMW between patients with sagittal synostosis after surgery and healthy children. Notably, PMW is not a surrogate for CI as the two measures vary independently. Instead, it appears to have an additive role, helping to describe where along the skull it is widest. While measuring PMW from a CT scan is more labor-intensive than direct anthropometric measurement of CI, it serves an adjunctive role for cephalometric analysis. In order to achieve better aesthetic results, the data suggest that physicians who now only measure CI to assess repair may benefit from measuring PMW as well, adjusting the reconstruction to normalize both measures.

Other groups have also realized the limitation in using cephalic index alone as a measure of aesthetic success after sagittal synostosis correction. Ruiz-Correa et al., (2006) published new scaphocephaly severity indices (SSIs) using CT scan bone windows to create specific two-dimensional planes from which to measure skull length and width.¹² When compared to the cephalic index, this study found that the SSIs had a higher specificity and sensitivity than cephalic index for quantifying sagittal synostosis.¹² Another study from the same institution also commented on the changes that occur regarding frontal bossing after surgical correction of sagittal synostosis, finding that frontal bossing decreased after open surgical correction by a combination of growth restriction as well as reduction.¹³ Marcus et al., (2006) created a mid-sagittal vector analysis (MSVA) to try to accurately define the distinctive head shape in sagittal synostosis patients.¹⁴ As a radial vector analysis, MSVA adequately captures the longitudinal differences associated with scaphocephaly, but does not address the transverse dimension.¹⁴ To our knowledge, our evaluation of PMW is the first study to define a new metric for assessing sagittal synostosis and compare it to normative data of healthy, matched controls.

One potential criticism of this study is the use of CT scans for postoperative evaluation and the additional radiation exposure to this patient population. Several centers routinely use CT scans to plan for operative procedures and evaluate surgical results after craniosynostosis operations. At our institution, we use a low-radiation-dose protocol with CT imaging. The studies are limited to a preoperative scan to analyze the brain, ventricle size, and suture fusion as well as a postoperative scan to evaluate for bone defects, overall shape, and cranial volume at approximately one year after surgery.⁵ In certain patients, even after surgery, skull growth is abnormal and the cephalic index may decline in the long-term.^{1, 11} A postoperative CT scan provides additional diagnostic information to diagnose regression and potentially plan for any secondary surgeries.

Future aims of this group will be to validate this measurement with a larger sample size to increase the power of our measurements. Additionally, we are also analyzing this measurement in a soft tissue window, which is more applicable to measurements made in clinical practice using calipers for direct anthropometry. Furthermore, as three-dimensional photography has been recently validated as a method to assess cranial volume comparable to the gold standard of CT scanning, we are also developing a similar protocol for measuring PMW from three-dimensional photographs.¹⁵ Finally, this metric has only been evaluated in patients who have undergone open surgical repair. Minimally-invasive techniques, such as endoscopic release, are becoming relatively popular for early intervention. Evaluation of PMW in patients undergoing other types of surgical repair may be beneficial.

In conclusion, this study introduces a novel measure (point of maximum width), which aids in quantifying the unique head shape associated with sagittal synostosis. As cephalic index generally increases after surgical correction, PMW also exhibits a consistent posterior shift with improvement in cranial shape. This metric can serve as a useful adjunct to cephalic index for evaluating the surgical outcomes of sagittal synostosis repair after open calvarial vault remodeling.

Acknowledgments

Disclosures: This work was supported by the St. Louis Children's Hospital Foundation – Children's Surgical Sciences Institute. Permission was obtained for use of patient images. This publication was made possible by Grant Number UL1 RR024992 from the National Center for Research Resources (NCRR), a component of the National Institutes of Health (NIH), and NIH Roadmap for Medical Research. Its contents are solely the responsibility of the authors and do not necessarily represent the official view of NCRR or NIH.

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Figure 1.
Normal CT scan in profile showing sella-nasion plane.

Figure 2. Normal skull, vertex view. Blue line indicates cranial length and green line indicates cranial width.

Figure 3.
Normal skull, vertex view. PMW calculated as blue line divided by sum of blue and red lines. $PMW = A/(A+B)$

Figure 4.
Point of maximum width in preoperative subjects and normal controls (age <12 months).

Figure 5.
Point of maximum width in postoperative subjects and normal controls (age >12 months).

Figure 6.
Average point of maximum width before and after surgical repair in cases and controls.

Figure 7.
Three patients with sagittal synostosis and normal cephalic index.

Table 1

Preoperative Assessment of Cranial Shape

	Sagittal Synostosis (preoperative) N = 27	Normal Controls N = 27	<i>p</i>
Point of Maximum Width	53% $\pm$ 1%	57% $\pm$ 1%	<0.001
Cephalic Index	66.8% $\pm$ 0.8%	83.3% $\pm$ 1%	<0.001

Table 2

Postoperative Assessment of Cranial Shape

	Sagittal Synostosis (postoperative) N = 14	Normal Controls N = 14	<i>p</i>
Point of Maximum Width	58% $\pm$ 1%	58% $\pm$ 1%	0.986
Cephalic Index	73.2% $\pm$ 1.1%	79.9% $\pm$ 1.4%	0.003