Impact of health disparities on treatment for single-suture craniosynostosis in an era of multimodal care

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Craniosynostosis is the premature fusion of the skull. There are two forms of treatment: open surgery and minimally invasive endoscope-assisted suturectomy. Candidates for endoscopic treatment are less than 6 months of age. The techniques are equally effective; however, endoscopic surgery is associated with less blood loss, minimal tissue disruption, shorter operative time, and shorter hospitalization. In this study, the authors aimed to evaluate the impact of race/ethnicity and insurance status on age of presentation/surgery in children with craniosynostosis to highlight potential disparities in healthcare access. Charts were reviewed for children with craniosynostosis at two tertiary care hospitals in New York City from January 1, 2014, to August 31, 2020. Clinical and demographic data were collected, including variables pertaining to family socioeconomic status, home address/zip code, insurance status (no insurance, Medicaid, or private), race/ethnicity, age and date of presentation for initial consultation, type of surgery performed, and details of hospitalization. Children with unknown race/ethnicity and those with syndromic craniosynostosis were excluded. The data were analyzed via t-tests and chi-square tests for statistical significance ($p < 0.05$). A total of 121 children were identified; 62 surgeries were performed open and 59 endoscopically. The mean age at initial presentation of the cohort was 6.68 months, and on the day of surgery it was 8.45 months. Age at presentation for the open surgery cohort compared with the endoscopic cohort achieved statistical significance at 11.33 months (SD 12.41) for the open cohort and 1.86 months (SD 1.1473) for the endoscopic cohort ($p < 0.0001$). Age on the day of surgery for the open cohort versus the endoscopic cohort demonstrated statistical significance at 14.19 months (SD 15.05) and 2.58 months (SD 1.030), respectively. A statistically significant difference between the two groups was noted with regard to insurance status ($p = 0.0044$); the open surgical group comprised more patients without insurance and with Medicaid compared with the endoscopic group. The racial composition of the two groups reached statistical significance when comparing proportions of White, Black, Hispanic, Asian, and other ($p = 0.000815$), with significantly more Black and Hispanic patients treated in the open surgical group. The results demonstrate a relationship between race and lack of insurance or Medicaid status, and type of surgery received; Black and Hispanic children and children with Medicaid were more likely to present later and undergo open surgery.

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CRANIOSYNOSTOSIS results in both cosmetic deformity and limitation in brain growth and development, requiring surgical intervention to avoid cognitive impairment and cosmetic dysmorphology. There are currently two principal forms of treatment for single-suture synostosis: open cranial vault remodeling and endoscopic suturectomy with postoperative orthotic therapy. Open surgery and endoscopic suturectomy are both effective means of treating craniosynostosis; however, the endoscopic approach is associated with decreased blood loss

and transfusion risk, decreased dissection and postoperative swelling, shorter operative time, and shorter hospitalization.¹ Candidates for endoscopic treatment are typically younger than 4 months of age at the time of surgery. Given the narrow age range for endoscopic treatment, early diagnosis is therefore essential to provide this option. As a result, for children with craniosynostosis to receive appropriate treatment, access to care and facilitation of diagnosis have been brought to the forefront.

It is well established that underrepresented minority

ABBREVIATIONS PCP = primary care physician.

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TABLE 1. Patient demographics

	No. of Patients	%
Total	121	100.0
Age at presentation, mos		
≤4	69	57.0
4–6	13	10.7
6–12	25	20.7
>12	14	11.6
Sex		
Male	82	67.8
Female	39	32.2
Race		
White	67	55.4
Black	10	8.3
Hispanic	25	20.7
Asian	10	8.3
Other	9	7.4
Surgical approach		
Open	62	51.2
Endoscopic	59	48.8

children are less likely to have regular access to health-care, have fewer physician visits, and have lower total healthcare expenditures compared with non-underrepresented minority children.² This trend is also apparent with regard to treatment for craniosynostosis. It has been demonstrated that African American children presented for initial consultation for craniosynostosis at an older age than their Caucasian counterparts.²

Given the benefits of endoscopic suturectomy and the prerequisite that patients eligible for this surgery must present before 4 months of age, the purpose of this study is to evaluate possible barriers to care experienced by minority patient populations. Since there is a true duality of treatment options for craniosynostosis at this time—open versus endoscopic repair, which is largely related to age at presentation—we specifically aimed to evaluate the impact of race/ethnicity and insurance status on age of presentation in children with single-suture craniosynostosis, thus highlighting potential disparities in healthcare access and care received by children with this condition and delineating targets for improvement in reducing these disparities.

Methods

A retrospective review of all children diagnosed with craniosynostosis who received care at NewYork-Presbyterian Weill Cornell Medical Center and Columbia University Medical Center/Morgan Stanley Children's Hospital of New York from January 1, 2014, to August 31, 2020, was conducted.

Periprocedural characteristics and outcomes were collected, including date of birth, race/ethnicity, age at initial consultation, date of initial consultation, diagnosis, date of surgery, age at surgery, location of surgery, miles from home address to surgical center, primary care physician

FIG. 1. Pie chart showing patients by race.

(PCP) name, PCP address, miles from home to PCP, insurance status (no insurance, Medicaid, or private), operation performed, length of hospitalization, complications, and need for revision surgery. Children with syndromic craniosynostosis or comorbidities were excluded from the data set; 49 children were excluded because their recorded race/ethnicity was “unknown” or “not recorded” (Table 1).

Statistical Analysis

The distribution of patient and procedural characteristics was evaluated, including frequency, mean, and standard deviations. Race/ethnicity was characterized as White, Black, Hispanic, Asian, or other. The data were analyzed using t-tests for age at presentation and age at surgery. Fisher's exact tests and chi-square tests were used to identify differences in the racial composition and type of insurance between the open and endoscopic surgical cohorts. Statistical significance was defined as $p < 0.05$.

Results

In this multiinstitutional, retrospective chart review, 121 children were identified with craniosynostosis who subsequently underwent surgical treatment. Of these 121 cases, 62 surgeries were performed in an open fashion (vertex craniectomy/cranial vault remodeling), and 59 were performed endoscopically (endoscope-assisted suturectomy).

Sixty-seven (55.4%) of the 121 patients identified as White, 10 (8.3%) identified as Black, 25 (20.7%) identified as Hispanic, 10 (8.3%) identified as Asian, and 9 (7.4%) identified as other (Fig. 1).

The mean age at initial presentation of the entire cohort was 6.68 months, and at the time of surgery it was

FIG. 2. Box plot showing the median age at presentation between the groups.

8.45 months. There was a statistically significant difference between patients undergoing open versus endoscopic surgery with regard to age at presentation; 11.17 months for the open cohort and 1.86 months for the endoscopic cohort ($p < 0.0001$) (Fig. 2). Similarly, there was a statistically significant difference in age at time of surgery for the open cohort versus the endoscopic cohort: 14.19 months for the former compared with 2.58 months for the latter ($p < 0.0001$) (Fig. 3).

With respect to insurance status, there was a significant difference between the two groups; 35 patients in the open surgery group were without insurance or with Medicaid, and 27 with private insurance, compared with 15 patients without insurance or with Medicaid in the endoscopic group, and 44 with private insurance (OR 3.802, 95% CI 1.701–7.998, $p = 0.0008$). The open surgery group comprised more patients without insurance or with Medicaid compared with the endoscopic group (Fig. 4): 56% of patients in the open surgery group had no insurance or Medicaid compared with only 25% of patients in the endoscopic group.

The racial composition of the two surgical groups was significantly different when comparing proportions of White, Black, Hispanic, Asian, and other ($\chi^2 = 19.84$, $p < 0.0001$). There were significantly more Black and Hispanic patients treated in the open surgery group (Fig. 5), where 25 (40.3%) of the 62 patients were White, 8 (12.9%) patients identified as Asian or other, and 29 (46.8%) patients identified as Black or Hispanic. In the endoscopic group, however, 42 (71.2%) of the 59 patients were White,

FIG. 3. Box plot showing the median age at surgery between the groups.

11 (18.6%) identified as Asian or other, and only 6 (10.2%) patients in this group were Black or Hispanic.

When evaluating patients by type of craniosynostosis, 52 (43.0%) of the 121 patients had sagittal synostosis, 39

FIG. 4. Bar graph showing insurance status versus surgery type. The blue bars indicate open surgery, and the orange bars indicate endoscopic surgery.

FIG. 5. Bar graph showing race versus surgery type. The green bars indicate open surgery, and the purple bars indicate endoscopic surgery.

(32.2%) had metopic synostosis, 26 (21.5%) had coronal synostosis (20 unilateral, 6 bilateral), and 4 (3.3%) had lambdoid synostosis (3 unilateral, 1 bilateral). Of the patients with sagittal synostosis, 24 (46%) of the 52 underwent open surgery, and 28 (54%) underwent endoscopic surgery. Of the 39 patients with metopic synostosis, 19 (49%) underwent open surgery, while 20 (51%) underwent endoscopic surgery. Of the patients with coronal synostosis, 16 (62%) underwent open surgery, while 10 (38%) underwent endoscopic surgery. Among those patients with lambdoid synostosis, 3 (75%) underwent open surgery and 1 (25%) had endoscopic surgery. Among the four types of synostosis, significantly more Black and Hispanic patients were treated with open surgery for the sagittal and metopic synostosis groups ($p = 0.0055$ and $p = 0.0059$, respectively) compared with White patients and patients who identified as Asian or other. In the coronal and lambdoid suture subtypes, no significant difference was noted between racial groups with regard to open versus endoscopic intervention.

Overall, there was a significant difference in age at presentation and surgical intervention when stratified by race (Fig. 6). The mean ages at presentation were 5.12 months for White patients, 9.16 months for Hispanic patients, and 14.45 months for Black patients ($p = 0.0364$). Surgery for craniosynostosis occurred at a mean age of 6.53 months for White children, 12.52 months for Hispanic children, and 16.65 months for Black children ($p = 0.0434$).

Discussion

While there have been a number of prior studies associating race and type of insurance with later presentation of children with craniosynostosis³⁻⁵ to a craniofacial specialist, to our knowledge, this is the first study of its

FIG. 6. Box plots showing the median age at presentation (A) and age at surgery (B) by race. Bar graph showing mean age by race (C). The yellow bars indicate the age at presentation, and the blue bars indicate the age at surgery.

kind to address how this discrepancy results in divergent treatment options in the current dual treatment paradigm of craniosynostosis (open vs endoscopic surgery) at institutions with no preference for either technique and where both techniques were used with equal frequency (49.6% vs 50.4%, respectively). Consistent with other studies addressing the impact of race on age of presentation, our results demonstrate that Black and Hispanic children were more likely to present later and to undergo open calvarial vault remodeling for sagittal and metopic synostosis than White children.

This disparity is especially important, as several prior studies have demonstrated that surgical correction of craniosynostosis within the 1st year of life, especially within the first 6 months of life, may be associated with better long-term cognitive outcomes as opposed to later surgery.^{6,7} Notably, this is an ongoing area of study with conflicting evidence; however, given the potential ramifications associated with inferior cognitive outcomes resulting from delayed surgical treatment, it is important to address. In a study of school-aged and adolescent children who had previously undergone surgical intervention for sagittal synostosis, patients treated before 6 months of age and those treated between 6 and 12 months were compared with those treated after 12 months. Patients who underwent surgery for sagittal synostosis before the age of 6 months demonstrated higher full-scale IQ and verbal IQ,⁷ word reading, reading comprehension, spelling, and numerical operations relative to patients who underwent surgery at a later age. Additionally, a higher percentage

of patients treated after 6 months of age had one or more reading-related learning disabilities compared with those with earlier surgery.⁷ These findings are echoed in a study by Bellew et al. comparing neurodevelopmental outcomes in patients who underwent cranial vault reconstruction before versus after 6 months of age. Again, it was noted that patients who underwent surgery before 6 months exhibited higher overall IQ, verbal IQ, reading comprehension, spelling, and numerical operations compared to those treated between 6 and 12 months of age and after 12 months.⁶

The results of our study demonstrate that Black and Hispanic patients presented later for initial consultation, and therefore underwent surgical intervention at an older age. These patients were subsequently more likely to undergo open surgery. Delayed presentation of minority children is also seen in other pediatric craniofacial abnormalities, such as cleft palate. In a study using the Pediatric Health Information System database, 2995 patients with cleft palate younger than 24 months of age were identified as having undergone cleft palate repair from 2003 to 2008. Age at repair was noted to be significantly delayed for patients without private insurance and patients of a non-White race/ethnicity compared to those with private insurance or White ethnicity.⁸

In a study published in 2008 in *Pediatrics* of 102,353 children, those of minority ethnicities/races were found to have visited pediatricians less frequently than White children.⁹ While this is a broad conclusion, this finding is consistent with the craniofacial literature where a 2017 paper by Gandolfi et al. determined that non-White patients were referred to a craniofacial specialist at an older age than their White counterparts. When evaluating patients by age of referral, only 27% were found to have been referred by the age of 3 months, which is the current Centers for Disease Control and Prevention consensus guideline for referral for craniosynostosis.¹⁰ Patients referred at younger than 3 months of age were more likely to be White, referred by a pediatrician (as opposed to self-referral or referral by another medical provider), and have an associated syndrome. Risk factors for very late presentation (> 12 months of age) were found to be non-White race/ethnicity, referral by someone other than a pediatrician, and multisuture involvement. In this study, there was no relationship between time to presentation and insurance status (private vs public).¹⁰

While there are clear benefits to endoscopic surgery for single-suture craniosynostosis—shorter operative and anesthesia times, shorter hospital stays, and lower rates and volumes of blood transfusions¹¹—other benefits include lower rates of reported stress among parents of children undergoing endoscopic surgery versus open surgery for craniosynostosis.¹²

The results of prior studies underscore the importance of early treatment for craniosynostosis, highlight the detrimental effects of delayed treatment, and reinforce the fact that later treatment can result in long-term inequities for the disproportionate number of minority and publicly insured children who receive delayed treatment. The findings of this study amplify the need to address barriers that may prevent Black and Hispanic patients, in addition

to those with no health insurance or public health insurance, from receiving early referrals to craniofacial specialists.

This study is limited by its retrospective nature in addition to patient self-reporting of race. Many patients in the original cohort were ultimately excluded as race was documented as “unknown,” likely due to inadequate documentation at the initial intake appointment, resulting in a smaller overall cohort size. Furthermore, when breaking down the cohort by type of synostosis, the sample size of the subgroups became quite small, which limits the generalizability of any conclusions made. Further study is warranted to determine whether distance traveled for care and distance from home to the PCP may also contribute to delayed presentation in children with single-suture craniosynostosis.

Conclusions

The results of this study demonstrate a clear relationship between race and lack of insurance or Medicaid status, and type of surgery received in children with craniosynostosis. Black and Hispanic children and children without insurance or with Medicaid were more likely to present later for evaluation of craniosynostosis, ultimately undergoing surgery at an older age. Many were thus ineligible for endoscopic surgery. Further research is needed to elucidate barriers to care for these at-risk groups, as well as potential solutions. Societal and institutional reforms are called for in order to reconcile these disparities in healthcare access.

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Disclosures

The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

Author Contributions

Conception and design: Imahiyero, Valenti. Acquisition of data: Imahiyero, Odigie, Warren. Analysis and interpretation of data: Valenti, Odigie. Drafting the article: Imahiyero, Hoffman, Valenti, Odigie. Critically revising the article: Imahiyero, Hoffman, Valenti. Reviewed submitted version of manuscript: Valenti, Premaratne.

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