A Retrospective Analysis of the Impact of Health Disparities on Treatment for Single Suture Craniosynostosis Before and During the Pandemic

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Abstract

Background: Candidates for endoscopic treatment for craniosynostosis must be less than 6 months old. Given the narrow window of eligibility, there is potential for barriers to access to impact the type of surgery a patient receives. We hypothesized that COVID may worsen these potential disparities.

Methods: Charts were reviewed for children with single suture craniosynostosis from January 2014 to March 2020 (pre-COVID cohort) and March 2020 to September 2021 (COVID cohort) from 2 academic centers.

Results: 110 children were in the pre-COVID group; 56 were treated open and 54 endoscopically. Of the 44 patients in the COVID cohort, 20 were treated endoscopically and 24 were treated open. There was no significant difference in age of presentation or age of surgery across cohorts when considering type of surgery performed. Rate of endoscopy in the pre-COVID versus COVID cohorts was not significantly different. In both the pre-COVID and COVID cohorts there were significant differences between age of presentation, age of surgery, and type of surgery received based on race—black and Hispanic patients presented later, had surgery later, and underwent open surgery more frequently than their white/Asian counterparts (P = .0095, P = .0067).

Conclusions: The pandemic did not alter age of presentation/age of surgery or rates of patients receiving endoscopic surgery in patients with single suture craniosynostosis. There was no relationship between insurance status and type of surgery received during the pandemic, however Hispanic and black patients presented later and underwent surgery at an older age compared to white/Asian patients as they did pre-pandemic.

Keywords

craniofacial morphology, pediatrics, epidemiology

Background

Craniosynostosis is the premature fusion of calvarial sutures resulting in both cosmetic deformity as well as limitations in brain growth and development. Surgical therapy is required to avoid cognitive impairment and cosmetic dysmorphology.

There are 2 principle means of treatment for single suture synostosis: open cranial vault remodeling and endoscopic suturectomy with post-operative orthotic therapy. While both approaches are currently accepted forms of therapy in the treatment of synostosis, endoscopic suturectomy is associated with decreased blood loss and transfusion risk, decreased dissection and post-operative swelling, shorter operative time, and shorter hospitalization.¹ Endoscopic therapy, however, is only successful in very young children, and candidates are typically less than 4 months of age at the time of surgery. Given the narrow age range for this treatment, early diagnosis is essential. Previous studies have demonstrated a delay in presentation and

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care for minority children and for those who are uninsured or publicly insured.²

Historically, we have learned that pandemics disproportionately affect poor and disadvantaged populations. Infectious disease transmission is accelerated by social determinants of health such as poor housing, overcrowding, and poor nutrition.³ The most recent COVID-19 pandemic has been no different, affecting minorities and those of lower socioeconomic status at higher rates. In a 2020 JAMA study performed in a large homeless shelter in Boston, of the 408 individuals living there, 147 or 36%, were found to have a positive SARS-CoV-2-PCR test, a rate much higher than the overall positivity rate in Massachusetts at that time (15.8%), likely due to the inability of these individuals to isolate.⁴ Regarding minority status, at the height of the pandemic, the COVID-19 infection rate was found to be 3 times higher in predominantly black counties than in white counties nationally; similarly, mortality was 6 times higher in black counties. In Chicago, over 50% of COVID cases and nearly 70% of COVID fatalities were within black cohorts even though blacks only comprise 30% of Chicago's population.⁵

Given the disproportionate toll of the pandemic on minorities and those of lower socioeconomic status, the purpose of this study was to evaluate the relationship of socioeconomic status and race/ethnicity on age of presentation and age of surgery in children with single suture craniosynostosis comparing cohorts before and during the pandemic to highlight the impact on disparities in health care received by children with this condition. We hypothesized that the pandemic would have widened the gap in health disparities in these patient populations and thus aimed to quantify the degree to which minority children and those of lower socioeconomic status

Table 1. Patient Demographics.

	Pre- COVID		COVID	
	No.	%	No.	%
Total	110	100.0	44	100.0
Age at presentation				
< 4 months	58	52.7	22	50.0
4-6 months	13	11.8	4	9.1
6-12 months	26	23.6	12	27.2
> 12 months	13	11.8	6	13.6
Sex				
Male	70	63.6	29	65.9
Female	40	36.4	15	34.I
Race				
White	60	54.5	24	54.5
Black	10	9.1	4	9.1
Hispanic	22	20.0	11	25.0
Asian	10	9.1	2	4.5
Other	8	7.2	3	6.8
Surgical approach				
Open	56	50.9	20	45.5
Endoscopic	54	49.1	24	54.5

were affected. To our knowledge, this is the first study of its kind to evaluate how the COVID pandemic affected disparities in care for children with craniofacial disorders.

Methods

Patients either self-referred or were referred to the craniofacial center by pediatricians, neurologists, ob-gyns, and neonatal intensive care physicians both within our hospital system and from the surrounding geographic area. All children with this condition were considered for inclusion however children with syndromic craniosynostosis or comorbidities were excluded from the data set. Children whose race/ethnicity was listed as "unknown" or "not recorded" were also excluded from analysis. All children were treated by a single craniofacial surgeon in conjunction with one of three pediatric neurosurgeons.

Peri-procedural 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, insurance status (no insurance, Medicaid, private insurance), operation performed, length of hospitalization, complications, type of suture involvement, and presence of any other medical conditions. For all patients, once diagnosis of craniosynostosis was made, efforts were made to minimize time between diagnosis and surgery by expeditiously pursuing medical clearance and booking patients for surgery as soon as possible for the patient and surgeon. All children in this cohort were considered to be candidates for endoscopic suturectomy with helmeting therapy independent of suture type if the diagnosis was made by age 4 months. Insurance status was demonstrated to be a proxy for socioeconomic status and children with no insurance or Medicaid presumed to be of lower socioeconomic status than those with private insurance.

Statistical Analysis

The distribution of patient and procedural characteristics was evaluated including frequency, mean, and standard deviations. Race/ethnicity was characterized as white, Hispanic, black, Asian, or other. The data were analyzed via T tests for age at presentation and age at surgery. Fisher's exact tests/Chi square tests were used to identify differences in the racial composition and type of insurance between the endoscopic and open surgical groups across the 2 cohorts (pre-COVID vs COVID). Statistical significance was defined as P < .05.

Results

In this multi-institutional, retrospective chart review, 154 total children were identified and treated for craniosynostosis of the coronal, sagittal, metopic, and lambdoid sutures. Of these 154, 110 children underwent surgery between January 2014 and February 2020 and were thus included in the "Pre-COVID cohort". Forty-four patients underwent treatment between

March 2020 and September 2021; these patients were included in the "COVID cohort" (Tables 1 and 2).

In the Pre-COVID cohort, 56 patients (50.9%) underwent open surgery while 54 underwent endoscopic treatment (49.1%). In the COVID cohort, 20 patients underwent open surgery (45.5%) while 24 underwent endoscopic treatment (54.5%) (Table 1). The proportion of patients undergoing open versus endoscopic therapy was not statistically different between the 2 groups (P = .59). Similarly, the proportion of patients identifying as white, black, Hispanic, Asian, and other were the same across the 2 time periods. Between the 2 time-defined cohorts, racial representation was not statistically different (Figure 1).

There was no significant difference found between the 2 cohorts in terms of Insurance Status. In the Pre-COVID cohort, 45 of the 110 patients (40.9%) had No Insurance or Medicaid, 65 patients (59.1%) had Private Insurance. In the COVID cohort 21 of the 44 patients had No Insurance or Medicaid (47.7%) while 23 (52.3%) had Private Insurance. In the Pre-COVID cohort among those without insurance or with Medicaid, 42.2% were white, 35.6% were Hispanic, 11.1% were black, 2.2% were Asian, and 8.9% identified as other. In the Pre-COVID cohort with private insurance, 63.1% were white, 9.2% were Hispanic, 7.7% were black, 13.8% were Asian and 6.2% identified as other. During the pandemic, among those without insurance or with Medicaid, 47.6% were white, 38% were Hispanic, 4.8% were black, 0% were Asian, and 9.5% identified as other. Those who presented during the pandemic and had private insurance 60.9% were white, 13.0% were Hispanic, 13.0% were black, 8.7% were Asian, and 4.3% identified as other (Table 3).

The average age of initial presentation of the entire Pre-COVID cohort was 6.35 months and 8.11 months at time of surgery compared to 6.00 months and 7.36 months, respectively, in the COVID cohort. There was no statistically significant difference between the 2 cohorts with regards to age of presentation or age at surgery (P=.82, P=.67). In patients who underwent endoscopic surgery, there was no significant difference between patients in the pre-COVID and COVID cohorts with regards to age at presentation (1.85 months vs 1.95 months, respectively [P=.75]). In terms of age at surgery in the endoscopic group, there was also no difference with average age of surgery 2.60 months in the

Table 2. Suture Type.

	Pre-COVID		COVID	
	No.	%	No.	%
Total	110	100.0	44	100.0
Type of synostosis				
Unicoronal	23	21%	11	25%
Metopic	38	34%	16	36%
Lambdoid	4	4%	2	5%
Sagittal	45	41%	15	34%

Pre-COVID cohort compared to 2.71 months in the COVID cohort (P = .65). Accordingly, there was no difference noted in lag time from presentation to surgery for endoscopic vs open surgery (0.75 months and 0.76 months, respectively). In patients who underwent open surgery, there was no significant difference between age of presentation or age at surgery in the Pre-COVID versus COVID groups (10.70 vs 10.85 months respectively, for age at presentation (P = .95), and 13.42 months and 12.95 months respectively, for age at surgery (P = .87)) (Figure 2).

With regards to insurance status, in the Pre-COVID cohort, there was a statistically significant relationship between insurance type and type of surgery whereby patients without insurance or with Medicaid presented later and were more likely to have undergone open surgery compared to endoscopic surgery. In the endoscopic cohort, 45 of 110 (40.9%) patients had Medicaid or were uninsured while 65 (59.1%) had Private Insurance. Among children with Medicaid/no insurance, 30 of 45 (66%) underwent open surgery compared to only 26 of 65 (40%) with private insurance (P = .0070). In the COVID cohort however, this relationship did not hold and there was no statistically significant difference between insurance status and type of surgery performed. In this group, 21 of 44 patients had Medicaid or were uninsured (47.7%) while 23 (52.3%) had Private Insurance. Among the children with Medicaid/no insurance, 9 of 21 (42.8%) underwent open surgery compared to 11 of 23 children (47.8%) with private insurance (P = .77) (Figure 3).

In the pre-COVID cohort, the racial composition of the 2 surgical groups was significantly different when comparing proportions of white, Hispanic, Asian, Black, and other (X^2) 16.96, [P=.0002]) with significantly more black and Hispanic patients treated in the open surgical group (Figure 4). In the open surgical group, 22 of the 56 patients were white (39.2%), 8 patients (14.3%) were Asian or other, and 26 (46.4%) identified as black or Hispanic. In the endoscopic group, however, 38 of the 54 patients were white (70%), 10 identified as Asian or other (18.5%), and only 6 patients (11.1%) identified as black or Hispanic. In the COVID cohort this relationship was no longer statistically significant; in the open surgical group 8 of the 20 patients were white (40%), 3 were black (15%), 7 were Hispanic (35%), 1 was Asian (5%), and 1 identified as other (5%). In the endoscopic group, 16 of 24 patients were white (66.7%), 1 was black (4.1%), 4 were Hispanic (16.7%), 1 was Asian (4.1%), and 2 identified as other (8.3%) (P = .13).

In both the Pre-COVID and COVID groups, when stratifying patients by race, there was a statistically significant difference in age of presentation and age at surgery with black and Hispanic patients presenting significantly later than their white counterparts. In the Pre-COVID cohort, age at presentation was 4.5 months for white patients, 14.45 months for black children, 9.11 months for Hispanic patients, 5.25 months for Asian patients, and 4 months for patients identifying as other; age at surgery was 5.80 months for white patients, 16.65 months for black patients, 11.97 months for Hispanic

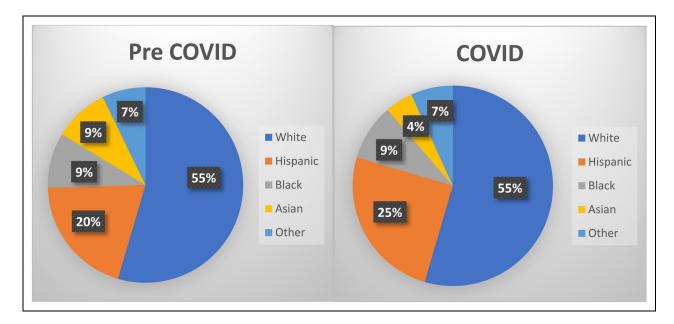


Figure 1. Racial demographics by cohort.

Table 3. Patient Insurance Status.

	Pre-COVID		COVID	
	No.	%	No.	%
Total	110	100.0	44	100.0
Insurance status				
No insurance/Medicaid	45	40.9	21	47.7
Private insurance	65	59.I	23	52.3
Medicaid/No insurance by race				
White	19	42.2	10	47.6
Black	5	11.1	I	4.8
Hispanic	16	35.6	8	38
Asian	I	2.2	0	0
Other	4	8.9	2	9.5
Private insurance by race				
White	41	63.I		60.9
Black	5	7.7		13.0
Hispanic	6	9.2		13.0
Asian	9	13.8		8.7
Other	4	6.2		4.3

patients, 6.65 for Asian patients, and 5.93 months for patients identifying as other (P = .016, P = .041, respectively). In the COVID cohort age at presentation was 3.55 months for white patients, 12.5 months for black patients, 9.63 months for Hispanic patients, 4.0 months for Asian patients, and 5.0 months for patients identifying as other; age at surgery was 4.65 months for white patients, 14.75 months for black patients, 11.54 months for Hispanic patients, 5.0 for Asian patients, and 5.5 months for patients identifying as other (P = .026, P = .010, respectively) (Figure 5).

When comparing racial groups across the 2 time cohorts, there was no significant difference in terms of age of presentation or age at surgery for each racial group indicating that average age of presentation and age at surgery remained consistent before and during the pandemic according to race.

Discussion

It is well documented that minority children and those of lower socioeconomic status have significant barriers to access in healthcare. In a 2007 study of 102,353 children, minority children were found to have visited pediatricians less frequently than Caucasian children.⁶ While this is a broad conclusion, this finding is consistent with the craniofacial literature where a 2017 paper by Gandolfi et al. determined that nonwhite 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 age 3 months, which is the current Centers for Disease Control (CDC) consensus guideline for referral for craniosynostosis.⁷ Patients referred at age less than 3 months were more likely to be white, referred by a pediatrician (as opposed to self-referral or referral by another medical provider), and to have an associated syndrome. Risk factors for very late presentation (over 12 months of age) were found to be nonwhite race/ethnicity, referral by someone other than a pediatrician, and multi-suture involvement. In this study, there was no relationship between time to presentation and insurance status (private vs public).⁷

Race and type of insurance have been associated with later presentation to a craniofacial specialist for children with craniosynostosis.^{2, 8–10} This study is the first of its kind to address the nature of this discrepancy during the COVID-19 pandemic.

Disparities related to delays in treatment in children with craniosynostosis are especially important to address as they

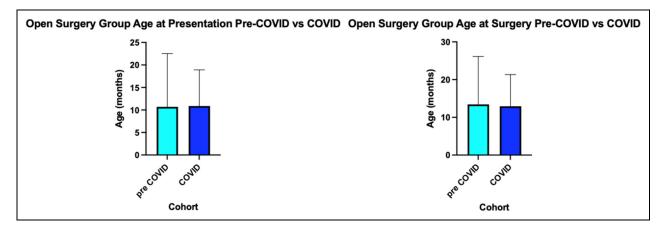


Figure 2. Age at surgery and age at presentation for patients undergoing open surgery for craniosynostosis.

may have significant clinical consequence. Surgical correction of craniosynostosis within the first year of life, especially within the first 6 months, may be associated with better longterm cognitive outcomes than later surgery.^{11, 12} While this is an ongoing area of study with some conflicting evidence, given the potential ramifications associated with inferior cognitive outcomes resulting from delayed surgical treatment, current trends are towards early intervention. In a study comparing neurodevelopmental outcomes in patients who underwent cranial vault reconstruction before and after 6 months of age, 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 of age.¹¹

Overall, the inequities in our healthcare system were exacerbated by the pandemic and children were disproportionately affected. Minorities and low income individuals were more likely to be essential workers and thus be unable to work remotely. This had an adverse effect on childcare and schooling which relied heavily on caregiver oversight. As such, school closures increased the gap in education between underrepresented minorities and those of low socioeconomic status and their white and higher economic status peers in addition to increasing food insecurity for children living in poverty who relied on school lunch programs.³

In medicine, this gap was similarly noted. In a study of pediatric ED utilization during the pandemic compared to prior, an overall decline in ED use across all races was noted with a more significant decrease noted in African American patients compared to non-Hispanic whites and in those with Medicaid or no insurance compared to those with private insurance.¹³ Given that minority race and Medicaid or non-insured patients are more likely to present to the ED for care as they lack other avenues, this implies an increase in healthcare disparities during the pandemic.¹³ Similarly, in a study from the pediatric otolaryngology department at Children's Hospital Los Angeles during COVID, the authors found that no show appointments were

more common in Hispanic patients and those who lived in a zip code that had a median income of less than 200% of the federal poverty level.¹⁴

In keeping with these known disparities, we hypothesized that we would see a greater delay in presentation and surgery in minority patients and in those with Medicaid/no insurance during the pandemic compared to before. While the results of our study do not demonstrate any widening of the already impressive gap in the dual treatment paradigm for craniosynostosis amongst different racial and socioeconomic groups, they continue to highlight the large degree of healthcare inequity. As we found in our previous study, in the COVID cohort, when stratified by race, patients of underrepresented minority status presented later and had surgery later compared to their white counterparts.

Despite this, race was not an independent predictor of open surgery in the COVID cohort. As the racial composition of the Pre-COVID and COVID cohorts were found to be similar, it may be deduced that at our institution, under represented minorities were presenting for care at similar frequencies compared to pre-pandemic levels. Still, black and Hispanic patients presented later for initial consultation and therefore underwent surgical intervention at an older age both before COVID and during the pandemic. In the COVID cohort, while age of presentation and age at surgery for black and Hispanic patients remained higher than white patients, white, black, Hispanic, Asian, and other patients were treated at equivalent frequencies in an open and endoscopic fashion unlike in the pre-COVID cohort where significantly more black and Hispanic patients were treated in the open surgical group compared to the endoscopic group. This discrepancy could be explained by sample size and the presence of outliers. As the time to surgery and time to presentation across all racial groups was lower during COVID (although not statistically significant), we could hypothesize that this may have contributed to this finding. While it would be convenient to presume that indeed, we have succeeded in narrowing the gap in disparities in our practice, the corollary would suggest that perhaps white children presented later due to a global decrease in access to care

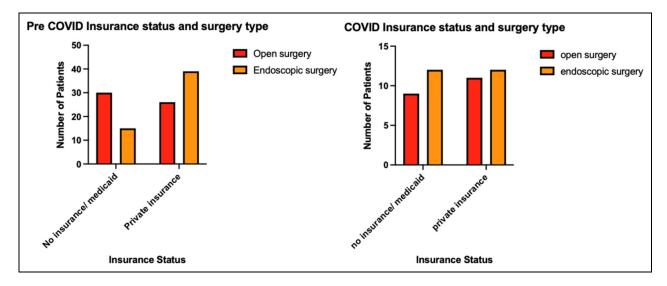


Figure 3. Relationship of insurance status to type of surgery.

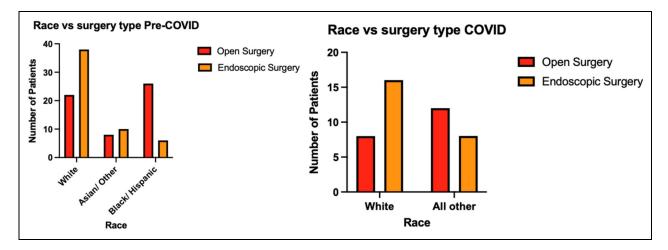


Figure 4. Relationship of race to type of surgery.

during the pandemic as well as increased video interfaces that may have missed early diagnoses.

The results of our study demonstrate that the proportion of patients undergoing open versus endoscopic therapy was not statistically different before and during the pandemic, indicating that both types of surgery were performed at similar frequencies. In conjunction with this finding, it was noted that age at presentation and age at surgery for the 2 time cohorts were the same, and even trended lower during the pandemic, indicating that COVID did not contribute to an overall delay in care and that consultations and surgeries were not generally postponed for children with this condition. Of note, our center has significantly increased in volume over the last few years as attending practices have become more established with more referring providers. This has been a direct result of coordinated efforts to increase practice access by launching new referral clinics within the community, establishing relationships with new referring providers, and allowing points of contact for patient self-referrals through the EMR.

With regards to insurance status, in the Pre-COVID cohort, there was a statistically significant relationship between insurance type and type of surgery as patients without insurance/ Medicaid presented later and were more likely to have undergone open surgery compared to endoscopic surgery. Interestingly, in the COVID cohort there was no statistically significant difference between insurance status and type of surgery performed, a small suggestion that our practice may be beginning to close the gap in disparities. Alternatively, this result may indicate an increased barrier to access during the pandemic in which more families switched from private insurance to Medicaid, or lost insurance entirely based on economic drivers and employment status.

The findings of this study amplify the need to address barriers that may prevent Black and Hispanic patients in addition

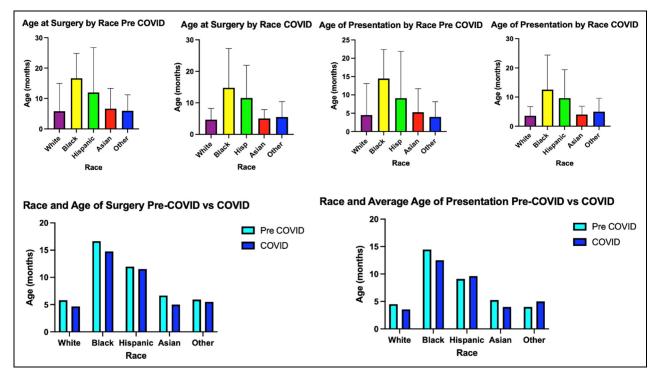


Figure 5. Age of surgery and age of presentation stratified by race.

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

While pre-existing disparities were generally upheld during the pandemic, in this specific patient population they were not exacerbated by COVID-19. One possible etiology of this may be that better served populations were also subjected to barriers to access with fewer pediatricians evaluating patients and holding virtual office hours. This may have resulted in delayed or missed diagnoses in previously well-served populations, which led to a decreased gap in time to presentation and time to surgery compared to pre-pandemic levels. Similarly, poorer populations and under-represented minorities were overall less able to practice social distancing and to utilize expensive technologies such as Telehealth which may have resulted in more in person visits and thus earlier detection of craniosynostosis. An important suggestion from this trend, therefore, may be to more thoroughly investigate the diagnostic sensitivity of video interfaces for craniofacial disorders, informing our practice moving forward. As many COVID-era practices will remain due to convenience and ease of access, our results may indicate an unintended ramification of these practices and indicate the need to transition back to exclusively in-person assessment for craniosynostosis.

Our results indicate that new challenges may have been introduced by the pandemic in terms of equality of care. Virtual assessments necessarily favor those with high speed internet, expensive devices to log onto appointments, and a quiet place to speak with a healthcare provider. Underserved patient populations often do not have the resources to engage in this type of care and this may perpetuate healthcare disparities in the future. More study is needed to evaluate the long-term effects of telemedicine on healthcare disparities.

This study has several limitations. It is retrospective in nature and given the short nature of the COVID cohort interval, sample size is small. Similarly, our study presumes that the long-term differences and results across the 2 surgical methods are equivalent. As this is not definitively validated at this time, this does remain a presumption. Despite this, the results demonstrate a clear relationship between delayed presentation and surgery in minority children and those with Medicaid/no insurance diagnosed with craniosynostosis. Further research is needed to elucidate specific barriers to care for these at risk groups, and most significantly, more work is needed to establish potential solutions. Systemic and institutional reforms are called for to reconcile these disparities in healthcare access.

Conclusions

The pandemic has reinforced the need to prioritize social determinants of health when implementing public health policies; COVID-19 has only served to highlight the profound disparities within our healthcare system. We are obligated to address these challenges.

Authors' Note

A retrospective review of all children diagnosed with craniosynostosis who received care at New York-Presbyterian/Komansky Children's Hospital and Columbia University Medical Center/Morgan Stanley Children's Hospital of New York from two separate epochs, January, 2014 to February 2020 and March 2020 to September 2021, was conducted (IRB number: 1206012507).

Declaration of Conflicting Interests

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