Sociodemographic disparities in fetal surgery for myelomeningocele: a single-center retrospective review

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OBJECTIVE Fetal surgery for myelomeningocele has become an established treatment that offers less risk of requiring a ventricular shunt and improved functional outcomes for patients. An increasing body of literature has suggested that social determinants of health have a profound influence on health outcomes. The authors sought to determine the socioeconomic and racial and ethnic backgrounds of patients who were treated with fetal surgery versus those who underwent postnatal repair.

METHODS Demographic data, the method of myelomeningocele repair, insurance status, and zip code data for patients entered into the National Spina Bifida Patient Registry (NSBPR) from Children’s Wisconsin were collected. The zip code was used to determine the Distressed Communities Index (DCI) score, a composite socioeconomic ranking with scores ranging from 0 (no distress) to 100 (severe distress). The zip code was also used to determine the median household income for each patient based on the US Census Bureau 2013–2017 American Community Survey 5-year estimates.

RESULTS A total of 205 patients were identified with zip code and insurance data. There were 23 patients in the fetal surgery group and 182 patients in the postnatal surgery group. All patients were born between 2000 and 2019. Patients in the fetal surgery group were more likely to have commercial insurance (100% vs 52.2%, p < 0.001). Fetal surgery patients were also more likely to be non-Hispanic White (95.7% vs 68.7%, p = 0.058), just missing the level of statistical significance. Patients who underwent fetal surgery tended to reside in zip codes with a higher median household income (mean $66,507 vs $59,133, p = 0.122) and less-distressed communities (mean DCI score 31.3 vs 38.5, p = 0.289); however, these differences did not reach statistical significance.

CONCLUSIONS Patients treated with fetal surgery were more likely to have commercial insurance and have a non-Hispanic White racial and ethnic background. The preliminary data suggest that socioeconomic and racial and ethnic disparities may exist regarding access to fetal surgery, and investigation of a larger population of spina bifida patients is warranted.

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KEYWORDS spina bifida; myelomeningocele; fetal surgery; socioeconomics; health disparities; congenital
have a profound effect on health outcomes, quality of life, and mortality. A previous study showed that sociodemographic factors had a significant influence on continence and ambulatory status in a large cohort of patients with spina bifida. Identifying preventable health disparities is critical to improving the outcomes and quality of life for patients with chronic diseases such as spina bifida. We sought to determine the sociodemographic background of patients who underwent fetal surgery for spina bifida compared with those who underwent traditional postnatal surgery.

Methods

Data Collection
Since 2009, patients receiving care through the Multidisciplinary Spina Bifida Clinic at Children's Wisconsin have been included in the National Spina Bifida Patient Registry (NSBPR). The NSBPR is a typical disease-based patient registry, partially funded and managed by the Centers for Disease Control and Prevention. There are 24 clinics across the country that participate in data entry for the registry. Data are collected on a yearly basis through interviews and examinations by the multidisciplinary treatment team. The NSBPR variables are recorded and then abstracted and entered anonymously into the database system. Each participating clinic has access to its own population data.

In September 2019, the NSBPR database for Children's Wisconsin was queried for children with myelomeningocele born in the United States and its territories between 2000 and 2019. The Children's Wisconsin IRB approved this study. The following variables were abstracted: year of birth, sex, the method of myelomeningocele repair, primary and secondary insurance at the time of the query, residential zip code at the time of the query, and race and ethnicity. The zip code was used to determine the median household income for individual zip code tabulation areas using the US Census Bureau 2013–2017 American Community Survey 5-year estimates in February 2020 (https://data.census.gov/cedsci/). In February 2020, zip code was also used to determine the 2018 Distressed Communities Index (DCI) score, a composite socioeconomic ranking with scores ranging from 0 (no distress) to 100 (severe distress). The DCI is a product of the Economic Innovation Group, a bipartisan public policy organization. The DCI for each residential zip code is based on the following 7 components: level of education in the adult population, housing vacancy rate, unemployment, poverty rate, median income ratio, change in employment, and change in number of business establishments. This is an established metric increasingly used to determine the effect of community distress on medical risks and outcomes.

Statistical Analysis
Statistical analysis was performed using R version 3.6.2 (The R Project). For categorical variables, each category and its frequency were computed, 95% confidence intervals were calculated, and the chi-square test was used to examine the relationship between the categorical variables for the postnatal and fetal surgery groups. Continuous variables were analyzed using mean and standard deviation within each group and compared between groups using the t-test. The significance level was set at p = 0.05.

Results
Overall, 205 patients were identified, 23 who had undergone fetal surgery and 182 who had undergone postnatal surgery (Table 1). Sixteen patients were “entered not enrolled” in the registry. These patients are treated at our center, but their families have not consented to enrollment and data collection for the NSBPR and, therefore, they are not part of the 205 patients in this study. There was a significant difference in the mean age of the two groups at the time of data collection (9.99 years postnatal surgery vs 6.0 years fetal surgery, p = 0.002), likely due to the increased availability of fetal surgery following publication of the MOMS in 2011. Only 3 of the patients who underwent fetal surgery were born between 2000 and 2010. There was a similar sex distribution between the fetal surgery and postnatal surgery groups (female 47.8% vs 53.8%, p = 0.746).

There was a significant difference in insurance status between the two groups, with 100% of the fetal surgery group and only 52.2% of the postnatal group having primary commercial insurance (p < 0.001). The overall hospital commercial insurance rate in 2019 was 52% (personal communication, hospital leadership). Patients who had undergone fetal surgery were also more likely to have a non-Hispanic White racial and ethnic background; however, this just missed the level of statistical significance (95.7% vs 68.7%, p = 0.058). The median household income based on the residential zip code showed that patients who had undergone fetal surgery tended to reside in areas with greater income (mean $66,507 vs $59,133, p = 0.122) and also tended to reside in areas with less community distress (mean DCI score 31.3 vs 38.5, p = 0.289); however, these differences did not reach the level of statistical significance. The highest quartile of DCI scores (> 75) is represented by communities that are severely distressed. In the fetal surgery group, 13.0% of patients came from communities considered significantly distressed, whereas 19.3% of patients in the postnatal surgery group came from significantly distressed communities (Fig. 1). Of note, only 2 patients who had undergone postnatal surgery had a change in residential zip code over time in the NSBPR, both due to military service. All other residential zip codes were unchanged over time.

Discussion
This retrospective review of patients treated at a single, multidisciplinary spina bifida clinic showed a statistically significant difference in insurance status in patients who were treated with fetal surgery compared with those who were treated with postnatal surgery. All patients in the fetal surgery group had primary commercial insurance compared with only 52.2% of the postnatal surgery group. In addition, nearly all patients in the fetal surgery group had a non-Hispanic White racial and ethnic background compared with 68.7% of the postnatal surgery group, although this difference fell short of statistical significance.
Patients who had undergone fetal surgery tended to live in less-distressed communities and communities with higher household income, yet these findings did not rise to the level of statistical significance. While our data are limited by a small sample size, the sociodemographic disparities noted in this preliminary study suggest that further study is warranted.

Publication of the MOMS trial in 2011 marked a turning point in the care of children with myelomeningocele. This cohort of patients has been the subject of longitudinal studies over the last several years. Many of the improvements noted in the prenatal surgery group have been sustained as the children have become school aged. While the cohort of patients in the MOMS informs much of our knowledge about open fetal surgery for myelomeningocele, little attention has been paid to the sociodemographic characteristics of the study population. In the MOMS, 94% of the mothers in the prenatal surgery group and 92% of the mothers in the postnatal surgery group were White. Greater than 90% of the trial popul-

<table>
<thead>
<tr>
<th>Characteristic</th>
<th>Fetal Surgery (n = 23)</th>
<th>Postnatal Surgery (n = 182)</th>
<th>p Value</th>
</tr>
</thead>
<tbody>
<tr>
<td>Sex, n (%) [95% CI]</td>
<td></td>
<td></td>
<td>0.746</td>
</tr>
<tr>
<td>F</td>
<td>11 (47.8) [26.8–68.4]</td>
<td>98 (53.8) [46.3–61.2]</td>
<td></td>
</tr>
<tr>
<td>M</td>
<td>12 (52.2) [30.6–73.2]</td>
<td>84 (46.2) [38.8–53.7]</td>
<td></td>
</tr>
<tr>
<td>Mean age, yrs (SD)*</td>
<td>6.00 (5.28)</td>
<td>9.99 (5.15)</td>
<td>0.002</td>
</tr>
<tr>
<td>Race &amp; ethnicity, n (%) [95% CI]</td>
<td></td>
<td></td>
<td>0.058</td>
</tr>
<tr>
<td>Hispanic</td>
<td>1 (4.3) [0.1–21.9]</td>
<td>28 (15.4) [10.5–21.5]</td>
<td></td>
</tr>
<tr>
<td>Non-Hispanic African American</td>
<td>0 (0.0) [0–14.8]</td>
<td>22 (12.1) [7.7–17.7]</td>
<td></td>
</tr>
<tr>
<td>Non-Hispanic White</td>
<td>22 (95.7) [78.1–99.9]</td>
<td>125 (68.7) [61.4–75.3]</td>
<td></td>
</tr>
<tr>
<td>Other</td>
<td>0 (0.0) [0–14.8]</td>
<td>7 (3.8) [1.6–7.8]</td>
<td></td>
</tr>
<tr>
<td>Primary insurance, n (%) [95% CI]</td>
<td></td>
<td></td>
<td>&lt;0.001</td>
</tr>
<tr>
<td>Commercial</td>
<td>23 (100.0) [85.2–100]</td>
<td>95 (52.2) [44.7–59.6]</td>
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</tr>
<tr>
<td>Medicaid</td>
<td>0 (0.0) [0–14.8]</td>
<td>80 (44.0) [36.6–51.5]</td>
<td></td>
</tr>
<tr>
<td>Other</td>
<td>0 (0.0) [0–14.8]</td>
<td>7 (3.8) [1.6–7.8]</td>
<td></td>
</tr>
<tr>
<td>Unknown</td>
<td>0</td>
<td>0</td>
<td></td>
</tr>
<tr>
<td>DCI score</td>
<td></td>
<td></td>
<td>0.289</td>
</tr>
<tr>
<td>Mean (SD)</td>
<td>31.27 (29.87)</td>
<td>38.46 (30.76)</td>
<td></td>
</tr>
<tr>
<td>Unknown, n</td>
<td>0</td>
<td>11</td>
<td></td>
</tr>
<tr>
<td>Median household income by residential zip code</td>
<td>$66,507.35 ($21,123.30)</td>
<td>$59,133.01 ($17,762.90)</td>
<td>0.122</td>
</tr>
<tr>
<td>Unknown, n</td>
<td>0</td>
<td>12</td>
<td></td>
</tr>
</tbody>
</table>

*Age at the time of data collection.
tion mothers were married or living with their partner. The average lengths of maternal schooling were 14.8 years and 15.0 years in the prenatal and postnatal surgery groups, respectively.1 Insurance status was not reported. These data suggest a study population with a high degree of social stability that is racially and ethnically homogeneous and well educated. Similar sociodemographic characteristics are found in post-MOMS fetal surgery cohorts reported in the literature.12,13

In comparison, a review of more than 2000 patients enrolled in the NSBPR gives a sense of the typical sociodemographic characteristics of the population of myelomeningocele patients. This review showed that 64.5% of patients had a non-Hispanic White racial and ethnic background and only 44.7% of patients had primary commercial insurance.14 Further study of this patient population showed that factors such as insurance status and race and ethnicity played a role in determining health outcomes.8

It remains unknown whether patients with spina bifida from a more diverse sociodemographic background treated with fetal surgery would have the same sustained improvements as those observed in the MOMS cohort. While the number of fetal surgery centers dramatically increased after the publication of the MOMS results, the intervention still requires a significant investment of time and effort on the part of the mother and family. Many families must travel far from home to a fetal center for the workup, surgery, and postsurgical care. Following surgery, the patient will spend several nights in the hospital and 2 weeks on modified bedrest and have weekly follow-up at most fetal surgery centers.1,15 This is a significant commitment, often taking families away from income-producing labor and social support for weeks or months. Families must also factor in the expenses that will be incurred once the child is born. The first few years of life are frequently a financially challenging time for families of children with spina bifida.16

Pediatric neurosurgeons continue to enjoy a prominent role in our society, and this gives us the unique ability to advocate for our patient’s.17 At a local and regional level, fetal surgery centers would be wise to have resources available for families with economic instability and less-robust social support. Outreach to community primary care physicians and obstetricians may help to limit disparities in the provision of care for mothers who are pregnant with a child with spina bifida. While fetal surgery is an expensive procedure with great costs to the healthcare system, the potential long-term improvement in outcomes for patients treated with fetal surgery may make the early investment worthwhile.18

There are a number of limitations to this study. As a single-center study, the number of patients, particularly in the fetal surgery cohort, is small. This limits the power of our conclusions. While there was a trend toward patients in the fetal surgery cohort residing in zip codes with higher median income and less community distress, these data did not rise to the level of statistical significance given the small sample size. Furthermore, all these metrics are imperfect markers of socioeconomic well-being. The NSBPR does not collect data on the level of parent education, parental occupation, or whether the parents of the child live together. Furthermore, while a large percentage of families in our spina bifida clinic choose to participate in the NSBPR, the registry does not represent all children with spina bifida treated at our center. At our most recent analysis of the institutional NSBPR data, 16 patients were not captured in the registry and not included in our study. Finally, between 2000 and 2010, the access to fetal surgery for patients was limited by the MOMS trial, which required travel to one of three centers and randomization to postnatal surgery for some of the patients included in the MOMS.

Conclusions

Our limited single-center data suggest that sociodemographic and racial and ethnic disparities exist regarding access to fetal surgery for myelomeningocele. Further study of the complete NSBPR is warranted, and efforts to mitigate healthcare disparities in this patient population should be pursued.

Acknowledgments

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References


**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: Foy. Acquisition of data: Foy, Derflinger, Sherburne. Analysis and interpretation of data: Foy, Sawin, Heffelfinger, Koop, Cohen, Sherburne. Drafting the article: Foy, Sherburne. Critically revising the article: Foy, Sawin, Heffelfinger, Koop, Cohen, Sherburne. Drafted submitted version of manuscript: Foy. Approved the final version of the manuscript on behalf of all authors: Foy. Study supervision: Foy.

**Supplemental Information**

**Previous Presentations**

This work was presented at the 49th Annual Meeting of the AANS/CNS Section on Pediatric Neurological Surgery, December 2–4, 2020, held virtually.

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