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CLINICAL ARTICLE

Socioeconomic factors associated with pediatric moyamoya disease hospitalizations: a nationwide cross-sectional study

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OBJECTIVE Healthcare disparities are widely described in adults, but barriers affecting access to care for pediatric patients with moyamoya disease (MMD) are unknown. Understanding socioeconomic factors impacting hospital access and outcomes is necessary to address pediatric healthcare disparities.

METHODS In this cross-sectional observational study, the Kids' Inpatient Database was used to identify patients admitted with a primary diagnosis of MMD from 2003 to 2016. Patients ≤ 18 years with a primary diagnosis of MMD based on *International Classification of Diseases* (ICD) codes were included. Hospital admissions were queried for use of cerebral revascularization based on ICD procedure codes.

RESULTS Query of the KID yielded 1449 MMD hospitalizations. After multivariable regression, Hispanic ethnicity (OR 0.52 [95% CI 0.33–0.81], p = 0.004) was associated with lack of surgical revascularization. Private insurance (OR 1.56 [95% CI 1.15–2.13], p = 0.004), admissions at medium- and high-volume centers (OR 2.01 [95% CI 1.42–2.83], p < 0.001 and OR 2.84 [95% CI 1.95–4.14], p < 0.001, respectively), and elective hospitalization (OR 3.37 [95% CI 2.46–4.64], p < 0.001) were positively associated with revascularization. Compared with Caucasian race, Hispanic ethnicity was associated with increased mean (± SEM) length of stay by 2.01 ± 0.70 days (p = 0.004) and increased hospital charges by $24,333.61 ± $7918.20 (p = 0.002), despite the decreased utilization of surgical revascularization. Private insurance was associated with elective admission (OR 1.50 [95% CI 1.10–2.05], p = 0.01) and admission to high-volume centers (OR 1.90 [95% CI 1.26–2.88], p = 0.002). African American race was associated with the development of in-hospital complications (OR 2.52 [95% CI 1.38–4.59], p = 0.003).

CONCLUSIONS Among pediatric MMD hospitalizations, multiple socioeconomic factors were associated with access to care, whether surgical treatment is provided, and whether in-hospital complications occur. These results suggest that socioeconomic factors are important drivers of healthcare disparities in children with MMD and warrant further study.

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KEYWORDS moyamoya disease; socioeconomic; healthcare disparities; vascular disorders

Moyamoya disease (MMD) is an intracranial arteriopathy characterized by progressive occlusion of the terminal internal carotid arteries and their main, proximal branches, as well as the development of atypical collateral vasculature. The posterior circulation may also be involved, although this is typically seen later in the disease course. MMD is a rare disorder, with an annual incidence ranging from 0.09 to 0.5 per 100,000 people.[1–4] It is a particularly severe cerebrovascular disease in children, with high rates of recurrent arterial ischemic stroke.[5,6] There is a female predominance for hospital admissions in both children and adults.[7,8] Historical data suggest a significantly higher incidence among Asian patients, but recent nationwide studies in the United States have demonstrated a disproportionately high incidence among African American patients.[1,7,8] This discrepancy

ABBREVIATIONS APR-DRG = All Patient Refined Diagnosis Related Group; ICD-CM = *International Classification of Diseases, Clinical Modification*; ICH = intracranial hemorrhage; KID = Kids' Inpatient Database; LOS = length of stay; MMD = moyamoya disease; PCS = Procedure Coding System; SCD = sickle cell disease.

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demonstrates the utility of large administrative databases to identify unique patient populations at risk for a given disease.

Socioeconomic factors are known to dramatically and pervasively impact the health and healthcare of children.[9] This is also true for patients in the fields of pediatric[10–15] and cerebrovascular[16–20] neurosurgery. Given the increasing clarity of the relationship between socioeconomic factors and health-related outcomes through the framework of biological, behavioral, physical environment, sociocultural environment, and systemic domains of influence,[21] and given the fact that structural forms of bias and racism, including access to the English language and access to health insurance and medical care, can impact individual health outcomes, it is important to further investigate the roles these factors play in the surgical management and in-hospital outcomes of children with MMD.

Recent studies have identified a number of socioeconomic risk factors associated with the incidence of MMD[1] and the impact of hospital volume on outcomes.[8] However, the impacts of socioeconomic factors on hospital admission, surgical revascularization, and outcomes in children with MMD have yet to be thoroughly investigated. In this study, our objective was to identify socioeconomic factors associated with MMD that may help in mitigating healthcare inequalities.

Methods

Study Cohort

This was a retrospective, cross-sectional study; data were obtained from the Kids' Inpatient Database (KID) for 2003–2016. The KID is a large, pediatric, inpatient database curated every 3–4 years by the Healthcare Cost and Utilization Project and created by the Agency for Healthcare Research and Quality; thus, KID data for the years 2003, 2006, 2009, 2012, and 2016 were included. Data included in the KID are based on administrative data created by hospitals. It includes pediatric admissions regardless of whether the facility is considered a children's hospital. Inclusion criteria for this study were age ≤ 18 years and a primary diagnosis of MMD by *International Classification of Diseases, Clinical Modification* (ICD-CM) code. Because of the nationwide transition from the *Ninth Revision* (ICD-9-CM) to the *Tenth Revision* (ICD-10-CM) in 2015, ICD-9-CM codes were used for data from 2003 to 2012, while ICD-10-CM codes were used for data from 2016. We noted whether the patient underwent cerebral revascularization during the hospitalization based on appropriate ICD-9 or ICD-10 Procedure Coding System (ICD-9-PCS or ICD-10-PCS, respectively) codes. The following ICD-9-CM/PCS codes were used: 437.5 (MMD); 430, 431, and 432 (intracranial hemorrhage [ICH]); and 39.28 (cerebral revascularization). The following ICD-10-CM/PCS codes were used: I67.5 (MMD); I60, I61, and I62 (ICH); and 031*0*G, where an asterisk denotes a position that could vary (cerebral revascularization). ICD codes were used to define in-hospital complications as described previously.[22] Institutional review board approval was not necessary for this publicly available database.

Demographic variables included age, sex, race/ethnicity, payer, All Patient Refined Diagnosis Related Group (APR-DRG) risk of mortality subclass, and income quartile. Income quartile reflects median household income for patient's zip code. Clinical variables included presence of ICH. Hospital variables included bed size and region. As previously described,[8,22] hospitals were categorized into low-, medium-, and high-volume centers based on the number of annual MMD admissions.

The primary outcome variable measured was whether hospital admissions included cerebral revascularization or not. Secondary outcome measurements included length of stay (LOS), hospital charges, and the development of any in-hospital complications.[22] Hospital charges were inflation-adjusted using price indexes for the gross domestic product from the US Department of Commerce Bureau of Economic Analysis. The adjustment used 2016 as the index base; that is, all charges are expressed in 2016 dollars.

Statistical Analysis

Patient discharges and charges were weighted using a weighting variable available within the database. Unadjusted odds ratios were calculated using univariable logistic regression. Multivariable binomial logistic regression with backward stepwise entry of variables was used to determine variables associated with cerebral revascularization and in-hospital complications. General linear models were used to determine variables associated with LOS and adjusted hospital charges. Variables with $p \leq 0.1$ in univariable analyses were included in the multivariable models. Given the high proportion of African American patients with a comorbid diagnosis of sickle cell disease (SCD), propensity score matching on age, APR-DRG risk of mortality, and SCD was performed to subsequently evaluate the association between African American race and in-hospital complications via logistic regression. As Healthcare Cost and Utilization Project databases do not contain unique patient identifiers, the data were not amenable to controlling for multiple measurements of a single patient. Missing data were excluded. Statistical significance was set at $p < 0.05$. Propensity score matching was performed in R (version 4.1.0, The R Project for Statistical Computing); all other analyses were performed using IBM SPSS Statistics (version 26.0, IBM Corp.).

Results

A total of 1449 hospital admissions met the inclusion criteria. Demographic and hospital-level characteristics are summarized in Table 1. The mean (± SEM) age at the time of admission was 9.0 ± 0.1 years, and there was a slight majority of female patients (54.8% vs 45.2% males). The plurality of admissions were for Caucasian patients (48.4%), followed by African American patients (20.2%), Hispanic patients (12.5%), and Asian or Pacific Islander patients (11.0%). Among the 249 hospitalizations for African American patients, 169 (67.9%) had a concomitant diagnosis of SCD. Private insurance was the primary payer in 52.5% of admissions, while Medicaid was the primary payer in 37.7%. The majority of admissions were elective (72.2%), and only 2.1% of admissions were associated with an ICH diagnosis. There were 440, 496, and 513 total hospital admissions for MMD at low-, medium-, and high-volume centers, respectively. The average number of MMD admissions per hospital was 2 at low-volume centers, 6 at medium-volume centers, and 22 at high-volume centers.

Cerebral Revascularization

Patient and hospital-level variables were evaluated for their association with cerebral revascularization (Table 2). Only 31.8% of hospitalizations for Hispanic patients included surgical revascularization compared with 49.2% for Caucasian patients, 40.2% for African American patients, 54.1% for Asian/Pacific Islander patients, and 49.5% for patients of other race/ethnicity ($p < 0.001$; Fig. 1A). Hispanic ethnicity (OR 0.52 [95% CI 0.33–0.81], p = 0.004) and admission to a hospital in the Midwest region (OR 0.58 [95% CI 0.37–0.92], p = 0.02) were associated with decreased odds of revascularization during a given hospital admission. On the other hand, private insurance (OR 1.56 [95% CI 1.15–2.13], p = 0.004), elective admission (OR 3.37 [95% CI 2.46–4.64], $p < 0.001$), admission to a hospital in the West region (OR 1.50 [95% CI 1.03–2.20], p = 0.04), and admission to medium-volume (OR 2.01 [95% CI 1.42–2.83], $p < 0.001$) or high-volume (OR 2.84 [95% CI 1.95–4.14], $p < 0.001$) centers were associated with increased odds of revascularization. Among the admissions for Hispanic patients, hospitalization at high-volume centers was associated with an increased rate of revascularization (61.5%) compared with hospitalization at low-volume (16.1%) and medium-volume (17.1%) centers ($p < 0.001$; Fig. 1B).

Patient Outcomes

Routine discharge disposition occurred in 1368 admissions (94.5%). The mean (± SEM) LOS for all admissions was 4.9 ± 0.2 days, and average adjusted hospital charges were $85,489.32 ± $2453.46. Hospital LOS for Hispanic patients was 7.4 ± 1.4 days compared with 4.7 ± 0.2 days for Caucasians, 4.6 ± 0.3 days for African Americans, 3.8 ± 0.2 days for Asian/Pacific Islanders, and 4.7 ± 0.4 days for other patients ($p < 0.001$; Fig. 1C). Adjusted hospital charges for Hispanic patients were $118,062.61 ± $10,419.01 compared with $89,471.31 ± $3801.16 for Caucasian patients, $69,929.49 ± $3875.66 for African American patients, $99,916.29 ± $8746.19 for Asian/Pacific Islander patients, and $79,748.26 ± $9694.48 for other patients ($p < 0.001$; Fig. 1D). After adjusting for age, sex, income, elective admission, hemorrhage, hospital MMD volume, hospital region, hospital bed size, payer, surgical revascularization during the hospitalization, and risk of mortality, Hispanic ethnicity was associated with an increased mean (± SEM) LOS of 2.01 ± 0.70 days (p = 0.004) and increased adjusted hospital charges of $24,333.61 ± $7918.20 (p = 0.002). There was no association between hospital MMD volume and LOS (p = 0.21) or adjusted charges (p = 0.08) among Hispanic patients. Payer status was not associated with LOS or hospital charges in the general linear models.

Given the socioeconomic factors associated with surgical revascularization and access to care, we also sought to

TABLE 1. Demographic details of pediatric admissions in the United States with a primary diagnosis code of MMD (2003–2016)

Characteristic	Value
Age, mean (SD), yrs	9.0 (4.8)
Sex, n (%)	
Male	654 (45.2)
Female	794 (54.8)
Race/ethnicity, n (%)	
Caucasian	595 (48.4)
African American	249 (20.2)
Hispanic	154 (12.5)
Asian or Pacific Islander	135 (11.0)
Other	97 (7.9)
Income quartile, n (%)	
≤25th percentile	307 (22.2)
25th–50th percentile	341 (24.7)
50th–75th percentile	341 (24.7)
>75th percentile	390 (28.3)
Payer, n (%)	
Medicaid	545 (37.7)
Private	758 (52.5)
Self-pay	35 (2.4)
Other	107 (7.4)
Elective admission, n (%)	
No	402 (27.8)
Yes	1043 (72.2)
APR-DRG risk of mortality subclass, n (%)	
Minor	1199 (82.7)
Moderate	133 (9.2)
Major	90 (6.2)
Extreme	27 (1.9)
ICH, n (%)	
No	1419 (97.9)
Yes	30 (2.1)
Hospital bed size, n (%)	
Small	108 (7.8)
Medium	532 (38.5)
Large	743 (53.7)
Hospital region, n (%)	
Northeast	345 (23.8)
Midwest	300 (20.7)
South	379 (26.2)
West	425 (29.3)
Hospital MMD vol, n (%)	
Low	440 (30.4)
Medium	496 (34.2)
High	513 (35.4)

Missing values are excluded. Percentages are reported for included values only.

TABLE 2. Predictors of surgical revascularization among pediatric admissions in the United States with a primary diagnosis code of MMD (2003–2016)

	No Revascularization	Revascularization	Unadjusted OR (95% CI)	Unadjusted p Value	Adjusted OR (95% CI)	Adjusted p Value
Age, mean (SD), yrs	9.1 (4.9)	9.0 (4.8)	1.00 (0.97–1.02)	0.69		
Sex, n (%)				0.68		
Male	376 (57.5)	278 (42.5)	Ref			
Female	448 (56.4)	346 (43.6)	1.05 (0.85–1.29)	0.68		
Race/ethnicity, n (%)				<0.001		**0.02**
Caucasian	303 (50.9)	292 (49.1)	Ref		Ref	
African American	149 (59.8)	100 (40.2)	0.70 (0.52–0.94)	0.02	1.17 (0.71–1.93)	0.55
Hispanic	105 (68.2)	49 (31.8)	0.48 (0.33–0.70)	<0.001	0.52 (0.33–0.81)	0.004
Asian or Pacific Islander	62 (45.9)	73 (54.1)	1.22 (0.84–1.78)	0.29	1.10 (0.70–1.72)	0.69
Other	49 (50.5)	48 (49.5)	1.02 (0.66–1.57)	0.93	0.75 (0.44–1.28)	0.29
Income quartile, n (%)				0.001		0.86
≤25th percentile	198 (64.5)	109 (35.5)	Ref		Ref	
25th–50th percentile	210 (61.6)	131 (38.4)	1.14 (0.83–1.57)	0.43	1.04 (0.70–1.54)	0.86
50th–75th percentile	194 (56.9)	147 (43.1)	1.39 (1.01–1.91)	0.04	0.97 (0.65–1.45)	0.87
>75th percentile	195 (50.0)	195 (50.0)	1.81 (1.34–2.48)	<0.001	1.13 (0.76–1.70)	0.55
Payer, n (%)				<0.001		**0.04**
Medicaid	364 (66.8)	181 (33.2)	Ref		Ref	
Private	388 (51.2)	370 (48.8)	1.93 (1.53–2.42)	<0.001	1.56 (1.15–2.13)	0.004
Self-pay	12 (34.3)	23 (65.7)	3.85 (1.88–7.87)	<0.001	2.03 (0.56–7.37)	0.28
Other	57 (53.3)	50 (46.7)	1.77 (1.17–2.69)	0.007	1.35 (0.79–2.30)	0.28
Elective admission, n (%)				<0.001		**<0.001**
No	324 (80.6)	78 (19.4)	Ref		Ref	
Yes	499 (47.8)	544 (52.2)	4.56 (3.46–6.00)	<0.001	3.37 (2.46–4.64)	<0.001
SCD, n (%)				0.03		0.33
No	682 (55.6)	544 (44.4)	Ref		Ref	
Yes	142 (63.7)	81 (36.3)	0.72 (0.54–0.97)	0.03	0.77 (0.46–1.30)	0.33
APR-DRG risk of mortality subclass, n (%)				0.007		0.37
Minor	661 (55.1)	538 (44.9)	Ref		Ref	
Moderate	79 (59.4)	54 (40.6)	0.84 (0.58–1.21)	0.34	1.15 (0.72–1.85)	0.55
Major	63 (70.0)	27 (30.0)	0.54 (0.34–0.86)	0.009	1.56 (0.84–2.87)	0.16
Extreme	21 (77.8)	6 (22.2)	0.35 (0.14–0.87)	0.02	0.59 (0.18–1.95)	0.38
ICH, n (%)				0.12		
No	803 (56.6)	616 (43.4)	Ref			
Yes	21 (70.0)	9 (30.0)	0.54 (0.24–1.19)	0.12		
Hospital bed size, n (%)				<0.001		0.22
Small	75 (69.4)	33 (30.6)	Ref		Ref	
Medium	236 (44.4)	296 (55.6)	2.87 (1.84–4.48)	<0.001	1.21 (0.66–2.20)	0.54
Large	466 (62.7)	277 (37.3)	1.36 (0.88–2.11)	0.17	0.91 (0.52–1.58)	0.73
Hospital region, n (%)				<0.001		**<0.001**
Northeast	189 (54.8)	156 (45.2)	Ref		Ref	
Midwest	221 (73.7)	79 (26.3)	0.44 (0.31–0.61)	<0.001	0.58 (0.37–0.92)	0.02
South	230 (60.7)	149 (39.3)	0.78 (0.58–1.05)	0.10	1.17 (0.78–1.74)	0.46
West	185 (43.5)	240 (56.5)	1.57 (1.18–2.09)	0.002	1.50 (1.03–2.20)	0.04
Hospital MMD vol, n (%)				<0.001		**<0.001**
Low	325 (73.9)	115 (26.1)	Ref		Ref	
Medium	288 (58.1)	208 (41.9)	2.06 (1.56–2.72)	<0.001	2.01 (1.42–2.83)	<0.001
High	211 (41.1)	302 (58.9)	4.07 (3.09–5.47)	<0.001	2.84 (1.95–4.14)	<0.001

Missing values are excluded. Percentages are reported for included values only. Adjusted odds ratios are provided only for variables entered into multivariable logistic regression. Boldface type indicates statistical significance for main categories in the adjusted analysis.

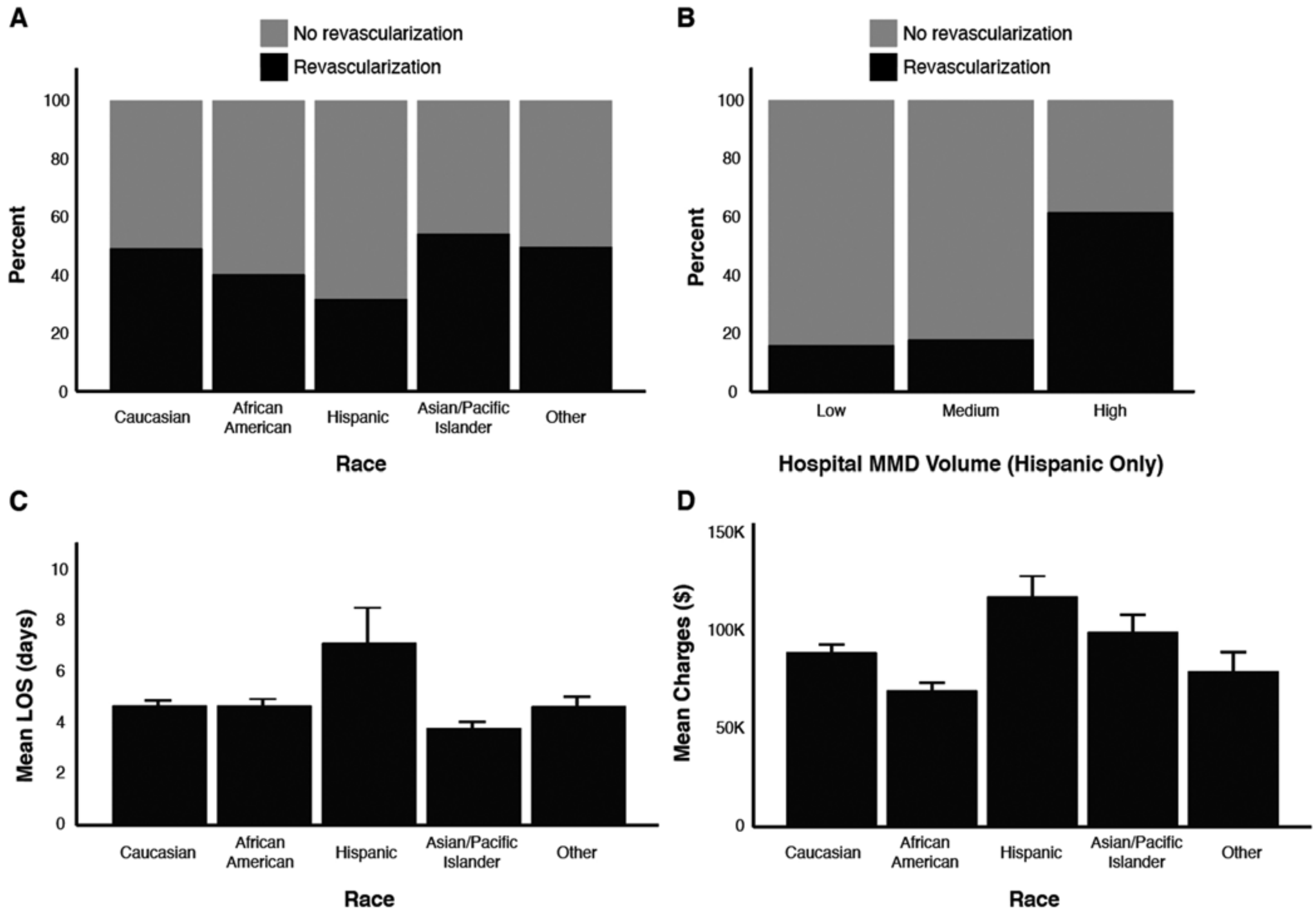

FIG. 1. Bar graphs showing that race is associated with surgical revascularization, LOS, and adjusted hospital charges. **A:** Percentage of admissions for each race category that were associated with surgical revascularization. **B:** Percentage of admissions for Hispanic patients that included surgical revascularization grouped by hospital MMD volume. **C:** Mean (± SEM) LOS for each race category. **D:** Mean (± SEM) adjusted hospital charges for each race category.

determine whether these factors were associated with the development of any in-hospital complications. Complications were noted in 9.2% of hospitalizations for African American patients compared with 4.5% for Caucasian patients, 3.9% for Hispanic patients, 3.7% for Asian/Pacific Islander patients, and 6.2% for other patients ($p = 0.05$; Fig. 2). Multivariable logistic regression revealed that African American race (OR 2.52 [95% CI 1.38–4.59], $p = 0.003$) and moderate, major, and extreme APR-DRG risk of mortality subclass ($p < 0.001$ for all) were associated with increased odds of complications (Table 3). The impact of African American race on development of complications was particularly related to surgical intervention; after adjusting for age, sex, income, elective admission, hemorrhage, hospital MMD volume, hospital region, hospital bed size, payer, and risk of mortality, African American race was associated with complications among hospitalizations utilizing cerebral revascularization (OR 4.17 [95% CI 1.73–10.02], $p = 0.001$) but not among hospitalizations without surgical intervention (OR 1.72 [95% CI 0.76–3.89], $p = 0.20$). Interestingly, of the 23 hospitalizations resulting in complications in African American pa-

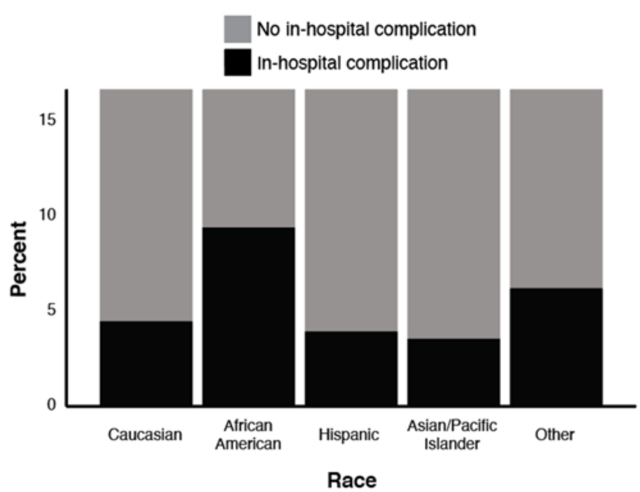

FIG. 2. Bar graph showing that race is associated with the development of in-hospital complications. Percentage of admissions for each race category that was associated with in-hospital complication.

TABLE 3. Predictors of any in-hospital complication among pediatric admissions in the United States with a primary diagnosis code of MMD (2003–2016)

	No Complication	Complication	Unadjusted OR (95% CI)	Unadjusted p Value	Adjusted OR (95% CI)	Adjusted p Value
Age, mean (SD), yrs	9.0 (4.8)	9.5 (5.3)	1.02 (0.97–1.07)	0.41		
Sex, n (%)				0.75		
Male	620 (94.8)	34 (5.2)	Ref			
Female	749 (94.3)	45 (5.7)	1.08 (0.68–1.70)			
Race/ethnicity, n (%)				0.04		0.01
Caucasian	568 (95.5)	27 (4.5)	Ref		Ref	
African American	226 (90.8)	23 (9.2)	2.22 (1.25–3.95)	0.007	2.52 (1.38–4.59)	0.003
Hispanic	148 (96.1)	6 (3.9)	0.88 (0.36–2.15)	0.77	0.73 (0.29–1.84)	0.51
Asian or Pacific Islander	130 (96.3)	5 (3.7)	0.79 (0.29–2.12)	0.64	0.89 (0.32–2.46)	0.82
Other	91 (93.8)	6 (6.2)	1.41 (0.57–3.52)	0.46	1.46 (0.57–3.73)	0.43
Income quartile, n (%)				0.82		
<25th percentile	290 (94.8)	16 (5.2)	Ref			
25th–50th percentile	320 (94.1)	20 (5.9)	1.14 (0.58–2.23)	0.70		
50th–75th percentile	325 (95.3)	16 (4.7)	0.87 (0.43–1.77)	0.70		
>75th percentile	366 (93.8)	24 (6.2)	1.16 (0.61–2.22)	0.66		
Payer, n (%)				0.99		
Medicaid	514 (94.3)	31 (5.7)	Ref			
Private	717 (94.6)	41 (5.4)	0.96 (0.59–1.56)	0.87		
Self-pay	33 (94.3)	2 (5.7)	0.81 (0.16–4.07)	0.79		
Other	101 (94.4)	6 (5.6)	1.02 (0.42–2.48)	0.97		
Elective admission, n (%)				0.04		0.96
No	372 (92.5)	30 (7.5)	Ref		Ref	
Yes	994 (95.3)	49 (4.7)	0.61 (0.38–0.97)	0.04	0.99 (0.55–1.750)	
APR-DRG risk of mortality subclass, n (%)				<0.001		**<0.001**
Minor	1158 (96.7)	40 (3.3)	Ref		Ref	
Moderate	113 (85.0)	20 (15.0)	5.14 (2.91–9.07)	<0.001	5.82 (3.10–10.91)	<0.001
Major	79 (87.8)	11 (12.2)	4.13 (2.06–8.29)	<0.001	4.97 (2.30–10.73)	<0.001
Extreme	20 (74.1)	7 (25.9)	10.51 (4.27–25.91)	<0.001	11.35 (4.08–31.59)	<0.001
ICH, n (%)				0.01		0.40
No	1344 (94.8)	74 (5.2)	Ref		Ref	
Yes	25 (83.3)	5 (16.7)	3.55 (1.32–9.55)	0.01	0.58 (0.16–2.06)	0.40
Hospital bed size, n (%)				0.90		
Small	102 (94.4)	6 (5.6)	Ref			
Medium	500 (94.0)	32 (6.0)	1.01 (0.42–2.44)	0.98		
Large	703 (94.6)	40 (5.4)	0.91 (0.38–2.15)	0.83		
Hospital region, n (%)				0.90		
Northeast	324 (93.9)	21 (6.1)	Ref			
Midwest	284 (94.7)	16 (5.3)	0.85 (0.44–1.67)	0.64		
South	360 (95.0)	19 (5.0)	0.78 (0.41–1.49)	0.45		
West	401 (94.4)	24 (5.6)	0.89 (0.49–1.63)	0.71		
Hospital MMD vol, n (%)				0.11		
Low	423 (96.1)	17 (3.9)	Ref			
Medium	470 (94.9)	25 (5.1)	1.32 (0.71–2.47)	0.38		
High	476 (92.8)	37 (7.2)	1.89 (1.05–3.39)	0.03		
Surgical revascularization, n (%)				0.90		
No	778 (94.4)	46 (5.6)	Ref			
Yes	591 (94.6)	34 (5.4)	0.97 (0.61–1.53)	0.90		

Missing values are excluded. Percentages are reported for included values only. Adjusted odds ratios are provided only for variables entered into multivariable logistic regression. Boldface type indicates statistical significance for main categories in the adjusted analysis.

tients, 16 (69.6%) involved a comorbid diagnosis of SCD. Additionally, of those 23 hospitalizations with associated complications, 12 occurred in the setting of surgical revascularization; in these surgical admissions, 11 (91.7%) involved a comorbid SCD diagnosis.

We sought to determine whether race or comorbid SCD was the primary driver of development of in-hospital complications given that 86.7% of hospitalizations involving a diagnosis of SCD were in African American patients. To do so, we used propensity score matching. Patients with and without in-hospital complications were matched for age, APR-DRG risk of mortality subclass, and SCD, resulting in 52 hospitalizations with complications and 52 without complications. After matching, univariable logistic regression indicated that African American race (compared with Caucasian race) was associated with the development of in-hospital complications (OR 2.29 [95% CI 1.00–5.47], p = 0.05).

Discussion

Socioeconomic factors commonly manifest as healthcare and health outcome disparities among pediatric patients. This finding has long been transparent on a global scale, but attention has recently turned to more subtle, underlying forms of health-related inequity that impact children in the United States. Socioeconomic factors have been shown to impact health even before birth, with African American and low-income pregnant women being disproportionately impacted by factors like regional air pollution, psychosocial stress, and suboptimal education levels, leading to undesirable outcomes such as preterm birth, low birth weight, and high infant mortality.[23] These associations remain after birth, with mortality rate disparities consistently found based on factors including race/ethnicity, education, income, access to a vehicle, and neighborhood. This holds true for all-cause mortality, as well as a wide variety of specific fields such as pediatric oncology, congenital cardiac surgery, trauma care, Down syndrome treatment, obesity management, and others.[9,24]

Unfortunately, cerebrovascular diseases are also impacted by socioeconomic disparities. Non-White race and insurance status have been associated with outcomes like worse access to treatment of unruptured aneurysms,[16,18] delayed time to carotid endarterectomy,[20] and increased LOS and higher mortality after acute ischemic stroke.[17]

Variables Associated With Cerebral Revascularization

In this nationwide analysis over multiple years, admission to medium- and high-volume centers, elective hospital admission, and private insurance were all independently associated with revascularization. Importantly, Titsworth and colleagues previously described that revascularization procedures performed at high-volume institutions were associated with improved LOS, cost, and mortality compared with those performed at low-volume centers, without an increase in complications.[8] This highlights the importance of concentrating MMD care at specialized, high-volume centers and underscores that access to such centers is paramount. Our study also revealed that rates of admissions including revascularization varied regionally across the United States; relative to the Northeast region, patients admitted to centers in the West were more likely to undergo revascularization, and those in the Midwest were less likely to undergo revascularization. The impact of regional and/or institutional practices regarding interventions for MMD may disproportionately affect those whose social and financial obligations leave them geographically confined to a region with low revascularization rates.

In addition, our results suggest that private insurance was independently associated with surgical revascularization, relative to Medicaid, potentially indicating a financial barrier to optimal treatment. Multiple prior studies have demonstrated the effect of race, income, or payer status on access to surgical intervention in adults, including for acute cholecystitis,[25] Parkinson disease,[26] and unruptured cerebral aneurysms.[16,18] A prior report by Kokoska et al.[27] demonstrated decreased access to laparoscopic cholecystectomy for pediatric appendicitis, and Rubinger et al.[13] showed that patients in low-income quintiles had a longer time to surgery than those in higher-income quintiles; however, to our knowledge, this is the first report to demonstrate that race or payer status was associated with surgical intervention during a given hospitalization in children with MMD. Of course, it is possible that payer status may reflect additional factors beyond family income, as children with chronic medical conditions may have public insurance regardless of parental income. We hope that our results can act as a springboard for more granular research in the future that may help clarify these nuances.

Interestingly, Hispanic ethnicity was independently associated with decreased odds of revascularization during a given hospitalization, although hospital admissions for Hispanic patients at high-volume centers were associated with revascularization more often than admissions at medium- or low-volume centers, further underscoring the importance of concentrating MMD care at specialized high-volume centers. Despite this, Hispanic ethnicity was associated with longer LOS and higher adjusted hospital charges compared with Caucasian race. To our knowledge, an association between Hispanic ethnicity and surgical revascularization for MMD has not previously been described. Although differences in MMD disease course and clinical phenotype at presentation have been described between Asian and non-Asian patients,[28–30] such differences have not been specifically described in Hispanic patients. As such, the fact that MMD admissions for Hispanic patients were less likely to include revascularization may demonstrate a genuine disparity in healthcare access and may be an opportunity for population-based measures aimed at mitigating this inequity. This interpretation is limited by the fact that the KID does not link patients across multiple hospitalizations. Thus, an alternative interpretation of these data is that Hispanic patients with MMD underwent revascularization at similar rates to other races but were admitted more often for other reasons. We believe that this alternative interpretation is unlikely, given the similarity in the top 4 secondary and tertiary diagnoses for MMD admissions in Hispanic patients. Hemiparesis/hemiplegia (n = 22, 21.0%), cerebral artery occlusion/stenosis or transient ischemic attack (n = 19, 18.1%), trisomy 21 (n = 15, 14.3%), and seizures/epilepsy (n = 13, 12.4%)

were the top 4 secondary and tertiary diagnoses among admissions that did not include revascularization (total of 105 admissions), whereas cerebral artery occlusion/stenosis or transient ischemic attack (n = 11, 22.4%), seizures/epilepsy (n = 9, 18.4%), hemiparesis/hemiplegia (n = 7, 14.3%), and trisomy 21 (n = 3, 6.1%) were the 4 most common secondary/tertiary diagnoses among admissions that included revascularization (total of 49 admissions). Even if the alternative interpretation were correct, however, we believe it would still reflect an important socioeconomic disparity in the disease course of MMD for Hispanic patients that should be further studied.

These results provide important insight into structural barriers to optimal MMD care, given that cerebral revascularization is associated with low annual stroke recurrence rates, especially when compared with patients not undergoing intervention.[31–33] The decreased access to revascularization may be due to a number of causes. First, it is highly likely that cultural differences influence individual families' health beliefs; thus, those differences may affect the likelihood of proceeding with surgical intervention. This may be compounded by language barriers during direct patient- or family-physician interactions and generally decreased health literacy.[34,35] Unfortunately, the KID does not provide granular data such as patient language that would allow us to differentiate whether such variables are associated with MMD hospitalizations. Second, it is well known that African American and Hispanic children are less likely to receive timely referrals to specialty care and obtain that medical care.[9] In fact, race was associated with admission to high-volume centers, with only 29.2% of hospitalizations for African Americans and 33.5% for Hispanics occurring at high-volume centers compared with 40.0% for Caucasians, 40.7% for Asian/Pacific Islanders, and 40.8% for other patients ($p < 0.001$). Although the exact reason is unknown, MMD hospitalizations for Hispanic patients are occurring more often at low- and medium-volume centers, and revascularization occurs less often at these centers. This is almost certainly compounded by minorities being less likely to have private insurance. We believe that these findings underscore the role that socioeconomic factors play in access to MMD treatment and hope that further studies will be performed to identify opportunities for public health intervention.

Socioeconomic Variables Associated With In-Hospital Complications

Clinical predictors of outcomes for MMD have been well described. Unfortunately, analyses of the racial/ethnic and socioeconomic factors associated with adverse events in this patient population are lacking. Our results suggest that African American race is independently associated with the development of in-hospital complications, particularly after surgical revascularization. Even among nonneurosurgical procedures, African American children have been found to have increased odds of complications after cholecystectomy[27] and cardiac transplantation[36] compared with Caucasian children. Furthermore, 91.7% of the surgical admissions of African American patients that resulted in complications included a comorbid SCD diagnosis. Studies have shown increased rates of perioperative morbidity for SCD patients after surgical intervention, including total hip arthroplasty[37] and renal transplantation.[38] Although this association may confound the finding that African American race is an independent predictor of perioperative complications in MMD, the results of our propensity score matching analysis suggest that race is independently associated with the development of in-hospital complications, even after matching for comorbid SCD. Ultimately, however, a randomized controlled trial would be necessary to definitively answer whether race or SCD predicts complications. Thus, we hope that this serves as a springboard for future research, especially as surgical revascularization for MMD patients with comorbid SCD has been described as an effective strategy to reduce the rates of recurrent stroke.[39–42]

Limitations

As with other studies utilizing administrative databases, this study has limitations. First, data entry relies on coding accuracy and cannot correct for errors in coding at the hospital level. Nevertheless, prior reports have estimated that, compared with medical records, ICD codes confer 89%–94% accuracy of diagnoses,[43–45] suggesting that they are reliable. In addition, to our knowledge, this study is the first to analyze pediatric MMD using the new ICD-10-CM coding system. While it is possible that there could be errors in converting ICD-9 to ICD-10 codes, given that moyamoya is coded as a single diagnosis in both systems, we believe that this series accurately represents the moyamoya patient cohort. Second, the ICD-9-PCS and ICD-10-PCS codes for cerebral revascularization do not significantly discriminate between direct and indirect revascularization. While our analysis could be confounded by choice of inclusion ICD codes, we attempted to minimize this by discussing all ICD-9-CM/PCS and ICD-10-CM/PCS codes with internal billing staff at our institution to best capture homogeneous populations across years. Third, as described above, the KID does not provide granular clinical information, including high-risk radiographic/angiographic factors, that could affect decisions to proceed with and outcomes related to revascularization. We attempted to control for this by including variables for risk of mortality in our multivariable models. Finally, the KID does not provide patient-specific data to link patients across multiple hospitalizations, and it does not provide long-term outcome assessments. Despite this, we believe that our findings identify important differences in access to care and hospital outcomes for MMD patients. Given its national lens, we believe the KID still provides valuable information about socioeconomic factors associated with pediatric healthcare.

Conclusions

Socioeconomic factors affecting access to care and outcomes for pediatric patients with MMD are lacking. In this nationwide cross-sectional study, race/ethnicity and socioeconomic determinants of treatment and outcome exist even after controlling for multiple patient- and hospital-level variables and after propensity score matching for comorbid SCD. These results highlight important pediatric healthcare disparities and provide guidance on areas for further study.

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References

1. Ghaffari-Rafi A, Ghaffari-Rafi S, Leon-Rojas J. Socioeconomic and demographic disparities of moyamoya disease in the United States. *Clin Neurol Neurosurg*. 2020;192:105719.
2. Kuriyama S, Kusaka Y, Fujimura M, et al. Prevalence and clinicoepidemiological features of moyamoya disease in Japan: findings from a nationwide epidemiological survey. *Stroke*. 2008;39(1):42-47.
3. Uchino K, Johnston SC, Becker KJ, Tirschwell DL. Moyamoya disease in Washington State and California. *Neurology*. 2005;65(6):956-958.
4. Wakai K, Tamakoshi A, Ikezaki K, et al. Epidemiological features of moyamoya disease in Japan: findings from a nationwide survey. *Clin Neurol Neurosurg*. 1997;99(suppl 2):S1-S5.
5. Fullerton HJ, Wintermark M, Hills NK, et al. Risk of recurrent arterial ischemic stroke in childhood: a prospective international study. *Stroke*. 2016;47(1):53-59.
6. McCrea N, Fullerton HJ, Ganesan V. Genetic and environmental associations with pediatric cerebral arteriopathy. *Stroke*. 2019;50(2):257-265.
7. Starke RM, Crowley RW, Maltenfort M, et al. Moyamoya disorder in the United States. *Neurosurgery*. 2012;71(1):93-99.
8. Titsworth WL, Scott RM, Smith ER. National analysis of 2454 pediatric moyamoya admissions and the effect of hospital volume on outcomes. *Stroke*. 2016;47(5):1303-1311.
9. Flores G. Technical report—racial and ethnic disparities in the health and health care of children. *Pediatrics*. 2010; 125(4):e979-e1020.
10. Attenello FJ, Ng A, Wen T, et al. Racial and socioeconomic disparities in outcomes following pediatric cerebrospinal fluid shunt procedures. *J Neurosurg Pediatr*. 2015;15(6):560-566.
11. Lin Y, Pan IW, Harris DA, Luerssen TG, Lam S. The impact of insurance, race, and ethnicity on age at surgical intervention among children with nonsyndromic craniosynostosis. *J Pediatr*. 2015;166(5):1289-1296.
12. Mukherjee D, Kosztowski T, Zaidi HA, et al. Disparities in access to pediatric neurooncological surgery in the United States. *Pediatrics*. 2009;124(4):e688-e696.
13. Rubinger L, Chan C, Andrade D, et al. Socioeconomic status influences time to surgery and surgical outcome in pediatric epilepsy surgery. *Epilepsy Behav*. 2016;55:133-138.
14. Stenzel AE, Fenstermaker RA, Wiltsie LM, Moysich KB. Disparities among racial/ethnic groups of patients diagnosed with ependymoma: analyses from the Surveillance, Epidemiology and End Results (SEER) registry. *J Neurooncol*. 2019; 144(1):43-51.
15. Walker CT, Stone JJ, Jain M, Jacobson M, Phillips V, Silberstein HJ. The effects of socioeconomic status and race on pediatric neurosurgical shunting. *Childs Nerv Syst*. 2014; 30(1):117-122.
16. Bekelis K, Missios S, Labropoulos N. Regional and socioeconomic disparities in the treatment of unruptured cerebral aneurysms in the USA: 2000-2010. *J Neurointerv Surg*. 2014; 6(7):556-560.
17. Fargen KM, Neal D, Blackburn SL, Hoh BL, Rahman M. Health disparities and stroke: the influence of insurance status on the prevalence of patient safety indicators and hospital-acquired conditions. *J Neurosurg*. 2015;122(4):870-875.
18. Guan J, Karsy M, Couldwell WT, et al. Factors influencing management of unruptured intracranial aneurysms: an analysis of 424 consecutive patients. *J Neurosurg*. 2017;127(1): 96-101.
19. Guha D, Ibrahim GM, Kertzer JD, Macdonald RL. National socioeconomic indicators are associated with outcomes after aneurysmal subarachnoid hemorrhage: a hierarchical mixed-effects analysis. *J Neurosurg*. 2014;121(5):1039-1047.
20. Wise ES, Ladner TR, Song J, et al. Race as a predictor of delay from diagnosis to endarterectomy in clinically significant carotid stenosis. *J Vasc Surg*. 2015;62(1):49-56.
21. NIMHD research framework. National Institute on Minority Health and Health Disparities; 2018. Accessed February 16, 2022. https://nimhd.nih.gov/about/overview/research-framework/nimhd-framework.html
22. Ravindra VM, Karsy M, Lanpher A, et al. A national analysis of 9655 pediatric cerebrovascular malformations: effect of hospital volume on outcomes. *J Neurosurg Pediatr*. 2019; 24(4):397-406.
23. Burris HH, Hacker MR. Birth outcome racial disparities: a result of intersecting social and environmental factors. *Semin Perinatol*. 2017;41(6):360-366.
24. Blakely T, Atkinson J, Kiro C, Blaiklock A, D'Souza A. Child mortality, socioeconomic position, and one-parent families: independent associations and variation by age and cause of death. *Int J Epidemiol*. 2003;32(3):410-418.
25. Greenstein AJ, Moskowitz A, Gelijns AC, Egorova NN. Payer status and treatment paradigm for acute cholecystitis. *Arch Surg*. 2012;147(5):453-458.
26. Chan AK, McGovern RA, Brown LT, et al. Disparities in access to deep brain stimulation surgery for Parkinson disease: interaction between African American race and Medicaid use. *JAMA Neurol*. 2014;71(3):291-299.
27. Kokoska ER, Bird TM, Robbins JM, Smith SD, Corsi JM, Campbell BT. Racial disparities in the management of pediatric appenciditis. *J Surg Res*. 2007;137(1):83-88.
28. Feghali J, Xu R, Yang W, et al. Racial phenotypes in moyamoya disease: a comparative analysis of clinical presentation and natural history in a single multiethnic cohort of 250 hemispheres. *J Neurosurg*. 2020;133(6):1766-1772.
29. Feghali J, Xu R, Yang W, et al. Differing surgical outcomes in a multiethnic cohort suggest racial phenotypes in moyamoya disease. *World Neurosurg*. 2019;128:e865-e872.
30. Hervé D, Ibos-Augé N, Calvière L, et al. Predictors of clinical or cerebral lesion progression in adult moyamoya angiopathy. *Neurology*. 2019;93(4):e388-e397.
31. Lee S, Rivkin MJ, Kirton A, deVeber G, Elbers J. International Pediatric Stroke S. Moyamoya disease in children: results from the International Pediatric Stroke Study. *J Child Neurol*. 2017;32(11):924-929.
32. Kuroda S, Hashimoto N, Yoshimoto T, Iwasaki Y. Radiological findings, clinical course, and outcome in asymptomatic moyamoya disease: results of multicenter survey in Japan. *Stroke*. 2007;38(5):1430-1435.
33. Cho WS, Kim JE, Kim CH, et al. Long-term outcomes after combined revascularization surgery in adult moyamoya disease. *Stroke*. 2014;45(10):3025-3031.
34. Baker DW, Parker RM, Williams MV, et al. The health care experience of patients with low literacy. *Arch Fam Med*. 1996;5(6):329-334.
35. Knapp C, Madden V, Marcu M, et al. Information seeking behaviors of parents whose children have life-threatening illnesses. *Pediatr Blood Cancer*. 2011;56(5):805-811.
36. Mahle WT, Kanter KR, Vincent RN. Disparities in outcome for black patients after pediatric heart transplantation. *J Pediatr*. 2005;147(6):739-743.
37. Fassihi SC, Lee R, Quan T, Tran AA, Stake SN, Unger AS.

Total hip arthroplasty in patients with sickle cell disease: a comprehensive systematic review. *J Arthroplasty*. 2020;35(8): 2286-2295.
38. Gérardin C, Moktefi A, Couchoud C, et al. Survival and specific outcome of sickle cell disease patients after renal transplantation. *Br J Haematol*. 2019;187(5):676-680.
39. Griessenauer CJ, Lebensburger JD, Chua MH, et al. Encephaloduroarteriosynangiosis and encephalomyoarteriosynangiosis for treatment of moyamoya syndrome in pediatric patients with sickle cell disease. *J Neurosurg Pediatr*. 2015; 16(1):64-73.
40. Hankinson TC, Bohman LE, Heyer G, et al. Surgical treatment of moyamoya syndrome in patients with sickle cell anemia: outcome following encephaloduroarteriosynangiosis. *J Neurosurg Pediatr*. 2008;1(3):211-216.
41. Newman S, Boulter JH, Malcolm JG, Pradilla I, Pradilla G. Outcomes in patients with moyamoya syndrome and sickle cell disease: a systematic review. *World Neurosurg*. 2020;135: 165-170.
42. Yang W, Xu R, Porras JL, et al. Effectiveness of surgical revascularization for stroke prevention in pediatric patients with sickle cell disease and moyamoya syndrome. *J Neurosurg Pediatr*. 2017;20(3):232-238.
43. Kokotailo RA, Hill MD. Coding of stroke and stroke risk factors using International Classification of Diseases, Revisions 9 and 10. *Stroke*. 2005;36(8):1776-1781.
44. Tirschwell DL, Longstreth WT Jr. Validating administrative data in stroke research. *Stroke*. 2002;33(10):2465-2470.
45. Berman MF, Stapf C, Sciacca RR, Young WL. Use of ICD-9 coding for estimating the occurrence of cerebrovascular malformations. *AJNR Am J Neuroradiol*. 2002;23(4):700-705.

Disclosures

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Conception and design: Raygor, Molinaro, Abla. Acquisition of data: Raygor, Phelps. Analysis and interpretation of data: Raygor. Drafting the article: Raygor, Phelps, Rutledge, Raper. Critically revising the article: all authors. Reviewed submitted version of manuscript: Abla, Raygor, Rutledge, Raper, Molinaro, Fox, Gupta. Statistical analysis: Raygor, Molinaro. Administrative/technical/material support: Abla. Study supervision: Abla.

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