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Scope of practice and outcomes of cerebrovascular procedures in children

Kimon Bekelis¹ · Symeon Missios² · Shannon Coy³ · Todd A. MacKenzie^{4,5,6,7}

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Abstract

Purpose The impact of scope of practice of providers (predominantly adult versus predominantly pediatric) on the outcomes of cerebrovascular procedures in children remains an issue of debate. We investigated the association of scope of practice with the outcomes of cerebrovascular interventions. *Methods* We performed a cohort study of all pediatric patients (younger than 18 years old) who underwent cerebrovascular procedures from 2009 to 2013 and were registered in the Statewide Planning and Research Cooperative System (SPARCS) database. In order to control for confounding, we used propensity score conditioning and mixed effects analysis to account for clustering at the hospital level.

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Kimon Bekelis kbekelis@gmail.com

- ¹ Section of Neurosurgery, Dartmouth-Hitchcock Medical Center, One Medical Center Drive, Lebanon, NH 03755, USA
- ² Department of Neurosurgery, Akron General Hospital, Akron, OH, USA
- ³ Department of Pathology, Brigham and Women's Hospital, Boston, MA, USA
- ⁴ The Dartmouth Institute for Health Policy and Clinical Practice, Lebanon, NH, USA
- ⁵ Department of Biomedical Data Science, Geisel School of Medicine at Dartmouth, Hanover, NH, USA
- ⁶ Department of Medicine, Dartmouth-Hitchcock Medical Center, Lebanon, NH, USA
- ⁷ Department of Community and Family Medicine, Dartmouth-Hitchcock Medical Center, Lebanon, NH, USA

Results During the study period, there were 1243 pediatric patients who underwent cerebrovascular procedures and met the inclusion criteria. Of these, 631 (50.7 %) underwent treatment by providers with predominantly adult practices and 612 (49.3 %) by physicians who operated predominantly on children. The mixed-effects multivariable regression analysis demonstrated lack of association of predominantly adult practice with inpatient mortality (OR, 1.20; 95 % CI, 0.61–2.38), discharge to a facility (OR, 1.50; 95 % CI, 0.73–3.09), and length of stay (LOS) (adjusted difference, 0.003; 95 % CI, -0.09 to 0.10). These associations persisted in propensity-adjusted models.

Conclusions In a cohort of pediatric patients undergoing cerebrovascular procedures from a comprehensive all-payer database, we did not demonstrate a difference in mortality, discharge to a facility, and LOS between providers with predominantly adult and predominantly pediatric practices.

Keywords Cerebrovascular intervention · Pediatrics · Scope of practice · SPARCS

Introduction

Cerebrovascular interventions are technically demanding, and their practice is typically limited to providers with additional fellowship training and particular subspecialty expertise [1, 6, 16, 17]. Most vascular pathology is encountered in adults. Although rare, pediatric cerebrovascular disease poses challenges related not only to the pathology but also to the patients' age and the specifics of clinical management in this population. In this case, the transition of care from adults to children is not intuitive. Pediatric patients have significantly smaller blood vessels, less atherosclerotic burden, higher frequency of syndromic pathology with multiple lesions, and



diseases that are extremely rare in adults, such as Moyamoya or vein of Galen malformations [3–5]. Pediatric neurosurgery requires extensive postresidency training to familiarize the physician with the specific challenges of this age group.

It is often questioned whether cerebrovascular specialists with adult practices can perform interventions in children as successfully and safely as providers focusing only on this age group do. However, limited literature exists attempting to answer this question. Prior studies of cerebrovascular interventions in children have reported the outcome of treating cerebral aneurysms, arteriovenous malformations (AVMs), and vein of Galen malformations and have demonstrated differences in characteristics and management in comparison to adults [3–5, 8–10, 15]. No prior study has investigated the association of scope of practice with the outcomes of cerebrovascular interventions in a comprehensive cohort of pediatric patients.

We performed a cohort study of pediatric patients, who were registered in the New York Statewide Planning and Research Cooperative System (SPARCS) [7], to investigate the association of predominantly adult practices with the outcomes of cerebrovascular interventions. The outcomes examined were inpatient mortality, length of stay, and discharge to a facility. We utilized a battery of approaches to control for confounding including regression adjustment, propensity score adjustment, and mixed effects models to account for clustering at the hospital level.

Methods

New York Statewide Planning and Research Cooperative System

All patients younger than 18 years who underwent cerebrovascular procedures and were registered in the SPARCS (New York State Department of Health, Albany, NY, USA) [7] database between 2009 and 2013 were included in the analysis. For these years, SPARCS contains patient-level details for every hospital inpatient stay and outpatient visit (including ambulatory surgery, emergency department services, and outpatient services rendered by hospital extension clinics) in the New York state as coded from admission and billing records. More information about SPARCS is available at https://www. health.ny.gov/statistics/sparcs/.

Cohort definition

We used the *International Classification of Disease-9-Clinical Modification* (ICD-9-CM) codes to identify patients with unruptured or ruptured cerebral aneurysms (ICD-9-CM code 437.3 or 430, respectively) who underwent clipping (ICD-9-CM code 39.51) or coiling (ICD-9-CM code 39.72,

39.75), craniotomy or endovascular approach for AVM (ICD-9-CM code 747.81 and 01.51, or 01.53, or 01.59, or 39.72), and bypass for Moyamoya disease (ICD-9-CM code (437.5 and 39.28) between 2009 and 2013. For patients with multiple interventions, only the first one was included in the final cohort.

Outcome variables

The primary outcome was inpatient mortality. Secondary outcomes were as follows: length of stay (LOS) during hospitalization and rate of discharge to a facility. Discharge to a facility was defined as discharge to any location other than the patient's home.

Exposure variables

The primary exposure variable was receiving treatment by a physician with a predominantly adult practice. A provider was considered to have a predominately pediatric cerebrovascular practice when more than 70 % of their total cerebrovascular case volume involved pediatric patients. Similarly, a provider was considered to have a predominantly adult practice when <70 % of their total cerebrovascular case volume involved pediatric patients and the majority of their patients were adults.

Treatment modality (clipping, coiling, craniotomy or endovascular approach for AVM, and bypass for Moyamoya) was included in the adjustment analysis. Data regarding attending-specific surgical volume (number of total cerebrovascular procedures) were also available and included in the adjustment.

The covariates (Table S1) used for risk-adjustment were age, gender, race (African-American, Hispanic, Asian, Caucasian, other), and insurance (private, Medicare, Medicaid, uninsured, other). The comorbidities used for risk adjustment were diabetes mellitus (DM), smoking, chronic lung disease, hypertension, hypercholesterolemia, congestive heart failure (CHF), history of stroke, transient ischemic attack (TIA), alcohol abuse, obesity, chronic renal failure (CRF), and coagulopathy. Only variables that were defined as "present on admission" were considered part of the patient's preadmission comorbidity profile.

Statistical analysis

To compare outcomes between providers with predominantly adult practices and providers with predominantly pediatric practices, we used several methods to address confounding, one of which was based on propensities. Initially, to compare inpatient mortality and discharge to a facility between predominantly adult practice providers and predominantly pediatric providers, we employed a multivariable logistic regression analysis including all the covariates listed above and the providers' case volumes. To account for the clustering of observations within hospital facilities, we employed mixedeffects models with facility name as a random-effect variable. For the analysis of LOS, we employed the corresponding versions of the multiple linear regression models. Due to the predominately positively skewed distribution of LOS, a logarithmic transformation of LOS was utilized.

To further control for confounding, we used regression models with adjustment (stratification) by deciles of propensity score. To derive the propensity of undergoing treatment by predominantly adult practice providers, we developed a prediction model using logistic regression based on the covariates described above.

Additionally, as part of the sensitivity analyses, we examined the effect of various other definitions of predominately pediatric cerebrovascular providers (in our primary analysis, those were defined as providers with more than 70 % of their practice consisting of pediatric patients) on outcomes [2]. Specifically, we repeated all the above analyses considering different thresholds to define predominately pediatric cerebrovascular providers, including providers with more than 90 or 100 % of their practice consisting of pediatric patients. However, due to the nature of the pathology and its rarity among pediatric populations, the above thresholds created significantly imbalanced cohorts. However, the direction of the observed associations was identical in these different iterations, and therefore these results are not reported. Lastly, in prespecified subgroup analyses, we examined the differences in outcomes for different procedures (clipping, coiling, craniotomy for AVM resection, bypass for Moyamoya), controlling for all covariates listed previously including case volume. The direction of the observed associations was identical in these subgroups, and therefore, these results are not reported.

Given that we had 1243 patients and approximately 50.3 % were treated by providers with predominantly adult practices, we had an 80 % power to detect a difference in mortality as small as 15.8 % between the two groups, at an α -level of 0.05. All probability values were the result of two-sided tests. Statistical analyses were performed using the 64-bit version of R.3.1.0 (R Foundation for Statistical Computing) and SPSS version 22 (IBM, Armonk, NY, USA).

Results

Patient characteristics

From 2009 to 2013, there were 1243 pediatric patients (average age 8.5 years, 48.7 % female) who underwent cerebrovascular interventions in the New York state and met the inclusion criteria for the study. From these patients, 631 (50.7 %) underwent treatment by a provider with predominantly adult practice, whereas 612 (49.3 %) underwent treatment by providers operating predominantly on children. The respective distribution of the exposure variables between the two types of providers can be found in Table 1.

Mortality

As demonstrated in Table 2, being treated by a predominantly adult practice provider was not associated with increased inpatient mortality (OR, 5.86; 95 % CI, 0.70–48.86) in the unadjusted analysis. Similarly, adjusting for confounders with a mixed-effects multivariable logistic regression model (Table 2) demonstrated a lack of association of predominantly adult practice with inpatient mortality (OR, 1.20; 95 % CI, 0.61–2.38), which persisted after the propensity score adjustment (OR, 5.81; 95 % CI, 0.67–50.49).

Length of stay

Among pediatric patients undergoing cerebrovascular procedures, the median LOS was 2 (IQR 3) after treatment by predominantly adult practice providers and 2 (IQR 2) after treatment by predominantly pediatric providers. As demonstrated in Table 2, being treated by a predominantly adult practice provider was associated with increased LOS (difference, 0.12; 95 % CI, 0.02-0.21) in the unadjusted analysis. However, adjusting for confounders with a mixed-effects multivariable logistic regression model (Table 2) demonstrated a lack of association of predominantly adult practice with LOS (adjusted difference, 0.003; 95 % CI, -0.09 to 0.10), which persisted after the propensity score adjustment (adjusted difference, 0.04; 95 % CI, -0.06 to 0.14).

Discharge to a facility

Among pediatric patients undergoing cerebrovascular procedures, 43 (6.9 %) patients were discharged to a facility after treatment by predominantly adult practice providers and 20 (3.3 %) after treatment by predominantly pediatric providers. As demonstrated in Table 2, being treated by a predominantly adult practice provider was associated with increased rate of discharge to a facility (OR, 2.20; 95 % CI, 1.28–3.78) in the unadjusted analysis. However, adjusting for confounders with a mixed-effects multivariable logistic regression model (Table 2) demonstrated a lack of association of predominantly adult practice with rate of discharge to a facility (OR, 1.50; 95 % CI, 0.73–3.09), which persisted after propensity score adjustment (OR, 1.73; 95 % CI, 0.98–3.07).

Table 1Patient characteristics

		Total pediatric patients $N = 1243$		Pediatric patients operated by surgeons with predominantly pediatric practice N=612		Pediatric patients operated by surgeons with predominantly adult practice N=631		-
		Mean	SD	Mean	SD	Mean	SD	P Value
Age		8.45	5.55	7.05	5.18	9.81	5.55	<0.0001
		Ν	%	Ν	%	Ν	%	
Female gender Race		605	48.67	291	47.55	314	49.76	0.435
Tuee	Caucasian	502	40.42	222	36.33	280	44.37	0.003
	African-American	104	8.37	34	5.56	70	11.09	0.0004
	Hispanic	162	13.04	69	11.29	93	14.74	0.070
	Asian	40	3.22	19	3.11	21	3.33	0.823
	Other	434	34.94	267	43.70	167	26.47	< 0.0001
Insurance								
	Medicare	0	0	0	0	0	0	-
	Medicaid	47	3.81	22	3.63	25	3.97	0.734
	Private	1088	88.10	551	90.92	537	85.37	0.009
	Uninsured	69	5.59	28	4.62	41	6.52	0.139
	Other	31	2.51	Ø	Ø	Ø	Ø	0.001
Procedure								
	Aneurysm clipping	14	1.13		Ø	Ø	Ø	0.309
	Aneurysm coiling	836	67.26	421	68.79	415	65.77	0.256
	AVM	368	29.61	172	28.10	196	31.06	0.253
	Moyamoya	25	2.01	Ø	Ø	Ø	Ø	0.494
Comorbidities								
	Ischemic stroke	Ø	Ø	Ø	Ø	Ø	Ø	Ø
	Lung disease	108	8.69	42	6.86	66	10.46	0.024
	Congestive heart failure	36	2.90	24	3.92	12	1.90	0.034
	Coagulopathy	16	1.29	Ø	Ø	Ø	Ø	0.951
	Chronic renal failure	Ø	Ø	Ø	Ø	Ø	Ø	0.309
	Hypertension	26	2.09	13	2.12	13	2.06	0.937
	Obesity	Ø	Ø	Ø	Ø	Ø	Ø	0.064

Based only on comorbidities present on admission. \emptyset means output suppressed to comply with the reporting rules of SPARCS, which do not allow printing of output involving less than 11 patients

SD standard deviation

Discussion

Using a comprehensive all-payer cohort of pediatric patients in the New York state who underwent cerebrovascular interventions, we did not identify an association of treatment by providers who hold a predominantly adult practice with inpatient mortality, LOS, or discharge to a facility. Our results were robust when considering several advanced observational techniques to account for confounders and control for clustering at the hospital level. The treatment of these patients can be demanding given the challenges of the pathology in this particular age group [3–5]. Little attention has been paid to the impact of the scope of practice of treating providers and the outcomes of this population.

Prior studies have examined the outcomes of cerebrovascular interventions in children [3–5, 8–10, 15]. Due to the rarity of this pathology in pediatric patients, most investigations are retrospective analyses of single-center experiences. Their results, therefore, have limited generalization. The lack of control for clustering and the limited use of multivariable methods additionally restrict the scope of these investigations. In one of the most recent series, Sanai et al. [13] summarized a

Model	Crude	Multivaria	ble regression adjusted ^a	Propensity score controlled		
	OR (95 % CI)	P value	OR (95 % CI)	P value	OR (95 % CI)	P value
Inpatient mortality ^b	5.86 (0.70-48.86)	0.102	1.20 (0.61–2.38)	0.598	5.81 (0.67–50.49)	0.111
Discharge to a facility ^b	2.20 (1.28-3.78)	0.004	1.50 (0.73-3.09)	0.265	1.73 (0.98-3.07)	0.06
	Difference (95 % CI)	P value	Difference (95 % CI)	P value	Difference (95 % CI)	P value
Length of stay ^c	0.12 (0.02–0.21)	0.017	0.003 (-0.09 to 0.10)	0.948	0.04 (-0.06 to 0.14)	0.425

Table 2 Association of receiving treatment by a predominantly adult practice neurosurgeon with outcome measures

OR odds ratio, 95 % CI 95 % confidence interval

^a Mixed effects, which includes treatment hospital as a random-effect variable

^b Analyses based on logistic regression

^c Analyses based on linear regression; point estimates are beta coefficients

30-year experience with cerebral aneurysm treatment in children for a single referral center. They were able to report on only 32 patients with 78 % good neurological recovery. The common denominator of all these experiences [3-5, 8-10, 15]is that pediatric cerebral aneurysms are more commonly giant or fusiform, posterior circulation based, and present with mass effect rather than subarachnoid hemorrhage. Additionally, mycotic aneurysms are more frequently observed in children in comparison to adults (10 versus 2-3 % respectively of all cerebral aneurysms) [3]. The analyses on the treatment of AVMs and vein of Galen malformations are not restricted to children and examine the multimodal treatment options available [4, 5]. No prior investigation in this population has focused on the practice characteristics of treating physicians, in an effort to reconcile the expertise of providers focusing on pediatric care and that of providers focusing on adults.

In our study of a broad range of cerebrovascular pathology, we intentionally addressed these limitations. First, we created a cohort of almost all pediatric patients in the New York state, giving a true picture of global practice in this age group. Our analysis for the first time considers the real-world representation of providers with predominantly adult practices, as well as predominantly pediatric providers to address this question. Second, we used advanced observational techniques to control for confounding including propensity score stratification. The possibility of clustering, which can bias the results of multicenter national studies, was accounted for at the hospital level. Results were consistent across techniques, supporting the validity of the observed associations.

Our study has several limitations common to administrative databases. First, residual confounding could account for some of the observed associations. However, this is minimized with the various advanced techniques we employed for risk-adjustment. Second, coding inaccuracies will undoubtedly occur and can affect our estimates. However, several reports have demonstrated that coding for cerebral aneurysms and cerebrovascular disease has shown nearly perfect association with medical record review [11, 14]. Additionally, coding for procedures is rarely inaccurate, given that it is a revenue generator, and is under scrutiny by payers.

Third, although SPARCS includes all hospitals from the entire New York state, the generalization of this analysis to the US population at large is uncertain. SPARCS does not provide any clinical information on the structure, size, or location of pathology, which are important factors in cerebrovascular interventions. However, we expect that physicians would treat similar pathology in a similar way regardless of whether they have a combined practice. Fourth, we were lacking long-term data on our patients. Quality metrics (i.e., modified Rankin score) are also not available through this database. Therefore, we cannot analyze the difference of physician expertise in regards to these measures. Alternatively, this question can be answered by the creation of large, long-term registries, with such efforts currently being underway [12]. Quality of life outcome measures or patient satisfaction metrics could be used instead in future prospective investigations. Finally, causality cannot be definitively established based on observational data, despite the use of advanced techniques.

Conclusions

The impact of the scope of practice of providers (predominantly adult versus predominantly pediatric) on the outcomes of cerebrovascular procedures in children remains an issue of debate. In a cohort of pediatric patients undergoing cerebrovascular procedures from a comprehensive all-payer database, we were not able to demonstrate a difference in mortality, discharge to a facility, and LOS between providers with predominantly adult practices and predominantly pediatric providers. Future comparative effectiveness studies will likely need to be based on prospective registries using quality outcome metrics. **Contributorship statement** KB performed the concept, design, manuscript preparation, and data interpretation.

SM performed the data analysis, statistical analysis, data interpretation, and critical review of the manuscript.

SC performed the data analysis, statistical analysis, data interpretation, and critical review of the manuscript.

TM performed the data analysis, statistical analysis, data interpretation, and critical review of the manuscript.

Compliance with ethical standards

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Competing interests There are no competing interests.

Data sharing All data are included in the study.

References

- Bekelis K, Goodney RP, Dzebisashvili N, Goodman DC, Bronner KK (2014) Variation in the care of surgical conditions: cerebral aneurysms. A Dartmouth Atlas of Health Care Series, Lebanon
- Bekelis K, Gottlieb D, Bovis G, Su Y, Tjoumakaris S, Jabbour P, MacKenzie TA (2015) Unruptured cerebral aneurysm clipping: association of combined open and endovascular expertise with outcomes. J Neurointerv Surg
- Blount JP, Oakes WJ, Tubbs RS, Humphreys RP (2006) History of surgery for cerebrovascular disease in children. Part I. Intracranial arterial aneurysms. Neurosurg Focus 20:E9
- Blount JP, Oakes WJ, Tubbs RS, Humphreys RP (2006) History of surgery for cerebrovascular disease in children. Part II. Vein of Galen malformations. Neurosurg Focus 20:E10
- Blount JP, Oakes WJ, Tubbs RS, Humphreys RP (2006) History of surgery for cerebrovascular disease in children. Part III. Arteriovenous malformations. Neurosurg Focus 20:E11

- Brinjikji W, Lanzino G, Kallmes DF, Cloft HJ (2014) Cerebral aneurysm treatment is beginning to shift to low volume centers. J Neurointerv Surg 6:349–352
- Health NYSDo (2015) Statewide Planning and Research Cooperative System (SPARCS). https://www.health.ny.gov/ statistics/sparcs/. Accessed 13 Feb 2015
- Herman JM, Rekate HL, Spetzler RF (1991–1992) Pediatric intracranial aneurysms: simple and complex cases. Pediatr Neurosurg 17:66–72
- Kalani MY, Elhadi AM, Ramey W, Nakaji P, Albuquerque FC, McDougall CG, Zabramski JM, Spetzler RF (2014) Revascularization and pediatric aneurysm surgery. J Neurosurg Pediatr 13:641–646
- Kim SK, Wang KC, Kim DG, Paek SH, Chung HT, Han MH, Ahn Y, Cho BK (2000) Clinical feature and outcome of pediatric cerebrovascular disease: a neurosurgical series. Childs Nerv Syst 16: 421–428
- Kokotailo RA, Hill MD (2005) Coding of stroke and stroke risk factors using international classification of diseases, revisions 9 and 10. Stroke 36:1776–17781
- NeuroPoint Alliance (2015) The National Neurosurgery Quality and Outcomes Database (N²QOD). http://www.neuropoint.org/ NPA N2QOD.html. Accessed 15 Jan 2015
- Sanai N, Caldwell N, Englot DJ, Lawton MT (2012) Advanced technical skills are required for microsurgical clipping of posterior communicating artery aneurysms in the endovascular era. Neurosurgery 71:285–294
- Tirschwell DL, Longstreth WTJ (2002) Validating administrative data in stroke research. Stroke 33:2465–2470
- Vanaman MJ, Hervey-Jumper SL, Maher CO (2010) Pediatric and inherited neurovascular diseases. Neurosurg Clin N Am 21:427– 441
- Zacharia BE, Bruce SS, Carpenter AM, Hickman ZL, Vaughan KA, Richards C, Gold WE, Lu J, Appelboom G, Solomon RA, Connolly ES (2014) Variability in outcome after elective cerebral aneurysm repair in high-volume academic medical centers. Stroke 45:1447–1452
- Zacharia BE, Ducruet AF, Hickman ZL, Grobelny BT, Badjatia N, Mayer SA, Berman MF, Solomon RA, Connolly ESJ (2011) Technological advances in the management of unruptured intracranial aneurysms fail to improve outcome in New York state. Stroke 42:2844–2849