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Factors associated with the timeliness of postnatal surgical repair of spina bifida

Elizabeth Radcliff[1], **Cynthia H. Cassell**[2], **Sarah B. Laditka**[3], **Judy K. Thibadeau**[2], **Jane Correia**[4], **Scott D. Grosse**[2], and **Russell S. Kirby**[5]

Elizabeth Radcliff: radclife@mailbox.sc.edu

[1]South Carolina Rural Health Research Center, Arnold School of Public Health, University of South Carolina, 220 Stoneridge Drive, Suite 204, Columbia, SC 29210, USA

[2]National Center on Birth Defects and Developmental Disabilities, Centers for Disease Control and Prevention, Atlanta, GA, USA

[3]Department of Public Health Sciences, College of Health and Human Services, University of North Carolina at Charlotte, Charlotte, North Carolina, USA

[4]Bureau of Epidemiology, Division of Disease Control and Health Protection, Florida Department of Health, Florida Birth Defects Registry, Tallahassee, Florida, USA

[5]Birth Defects Surveillance Program, Department of Community and Family Health, College of Public Health, University of South Florida, Tampa, Florida, USA

Abstract

Purpose—Clinical guidelines recommend repair of open spina bifida (SB) prenatally or within the first days of an infant's life. We examined maternal, infant, and health care system factors associated with time-to-repair among infants with postnatal repair.

Methods—This retrospective, statewide, population-based study examined infants with SB born in Florida 1998–2007, ascertained by the Florida Birth Defects Registry. We used procedure codes from hospital discharge records to identify the first recorded myelomeningocele repair (ICD-9 CM procedure code 03.52) among infants with birth hospitalizations. Using Poisson multivariable regression, we examined time-to-repair by hydrocephalus, SB type (isolated [no other coded major birth defect] versus non-isolated), and other selected factors.

Results—Of 199 infants with a recorded birth hospitalization and coded myelomeningocele repair, 87.9 % had hydrocephalus and 19.6 % had non-isolated SB. About 76.4 % of infants had repair by day 2 of life. In adjusted analyses, infants with hydrocephalus were more likely to have

Correspondence to: Elizabeth Radcliff, radclife@mailbox.sc.edu.

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Compliance with ethical standards The Institutional Review Boards at the University of North Carolina at Charlotte, the FDOH, and CDC approved this study.

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

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timely repair (adjusted prevalence ratio (aPR) = 1.48, 95 % confidence interval (CI) 1.02–2.14) than infants without hydrocephalus. SB type was not associated with repair timing. Infants born in lower level nursery care hospitals with were less likely to have timely repairs (aPR = 0.71, 95 % CI 0.52–0.98) than those born in higher level nursery care hospitals.

Conclusions—Most infants with SB had surgical repair in the first 2 days of life. Lower level birth hospital nursery care was associated with later repairs. Prenatal diagnosis can facilitate planning for a birth hospital with higher level of nursery care, thus improving opportunities for timely repair.

Keywords

Birth defects; Spina bifida; Pediatric surgery; Timeliness

Introduction

Spina bifida (SB) is a neural tube defect that results from a failure of the caudal neural tube to close early in embryonic development [22]. The recommended standard of treatment for open presentations of SB is prenatal surgical repair or postnatal repair within the first few days of life [4, 20]. If an infant does not undergo prenatal SB closure, prompt postnatal closure is essential to prevent infection and protect the exposed nerves from additional trauma [21, 23]. Prompt postnatal repair has been associated with reduced risk of ventriculoperitoneal shunt infection [14], neurogenic bladder [38], and neurodevelopmental delays [24].

Timely care among children with birth defects, such as SB, remains an understudied area [5, 27, 28, 42]. Previous relevant studies were based on 20-year-old data [9, 36], were descriptive only [9, 36], used a convenience or small clinical sample [9, 14, 36], or used only hospital discharge data to identify SB and demographics [9, 17].

Our study used a statewide, population-based sample of infants with SB to provide information on the timing of post-natal repair of SB. We hypothesized that the majority of infants would undergo surgical repair within the first 2 days of life. We also examined associations with selected maternal, infant, and health care system characteristics [19, 37, 40]. A focus of our analysis was whether disease severity, specifically hydrocephalus and non-isolated SB (SB with another coded major birth defect), was associated with timing of surgical repair.

Methods

Design and study population

This study was a retrospective, population-based observational study of infants with SB born in Florida between January 1, 1998 and December 31, 2007. We obtained the data from linked datasets provided by the Florida Birth Defects Registry (FBDR) and the Florida Bureau of Vital Statistics, both in the Florida Department of Health (FDOH), and from the Florida Agency for Health Care Administration (AHCA). We used the AHCA data from

January 1, 1998, through December 31, 2008, to ensure adequate hospital discharge data for each infant.

The FBDR is a statewide, population-based passive birth defects surveillance system. The FBDR identified infants with SB without anencephaly using the *International Classification of Disease, 9th revision; Clinical Modification* (ICD-9-CM) codes 741.00–741.93. The FBDR excludes infants who were adopted or prospectively adopted or born out of state. The FBDR includes infants if they are live-born in Florida to a mother who is a Florida resident at delivery [30, 31]. For this study, infants needed to match to an inpatient birth hospitalization discharge record. We excluded infants without a matched birth hospitalization to reduce error that could result from a repair occurring earlier in a hospital that did not report discharge data to the AHCA.

Following consultation with clinical experts from the Centers for Disease Control and Prevention's (CDC) National Center on Birth Defects and Developmental Disabilities (NCBDDD), we used the ICD-9-CM procedure codes 03.51 (meningocele (MC) repair) and 03.52 (myelomeningocele (MMC) repair) to identify surgical repair of SB. We further restricted the analysis to infants with a coded MMC repair (03.52). This restriction ensured a more uniform study population and more reliably captured infants with open SB presentations, which require prompt surgical repair.

Two infants who died during the first 28 days of life were excluded from analyses because they may have had more severe or complex medical conditions than infants who survived the neonatal period. Thus, their experience of surgical repair may not be typical of infants with SB. We retained infants who died later in infancy or during childhood to capture the full extent of factors associated with timely surgical repair among infants with SB who had a repair.

Variable construction

Outcome variable—The primary outcome of interest was timely postnatal surgical repair of the infant with SB. Using recommended guidelines for postnatal surgical repair [4, 20], infants were considered to have timely repair if the procedure code for the repair was on day 0, 1, or 2 of life. Surgical repair after this period was considered a later repair. If an infant had more than one repair procedure code listed, we used the time associated with the first recorded repair. We analyzed time-to-repair as a binary variable of ≤2 versus >2 days.

Exposure variables—The primary exposure of interest was SB type, isolated or non-isolated, which was coded as a dichotomous variable. Infants were classified as having isolated SB if they met any of the following criteria: (1) had only SB and no other coded major birth defect; (2) had only SB and a minor birth defect, such as low set ears or skin tags; or (3) had only SB accompanied by a documented sequence of birth defects related to SB, such as hydrocephalus, and no additional unrelated major birth defects [6, 18, 26]. Classification of isolated or non-isolated SB (hereafter, SB type) was informed by previous research and in consultation with CDC clinical experts [6, 18, 26].

A second exposure of interest was the presence of hydrocephalus, coded as a dichotomous variable. Following consultation with clinical experts from CDC's NCBDDD, we identified hydrocephalus using ICD-9-CM codes 741.01–741.03. We expected that the presence of hydrocephalus would serve as a proxy for more severe forms of SB, such as MMC, based on the high percentage of infants with MMC who also have hydrocephalus [8, 21]. The ICD-9-CM diagnostic codes for SB in administrative data do not differentiate between MMC and other cases.

Covariates—The selection of covariates was informed by previous research on timeliness of care or access to care among children with special health care needs (CSHCN), including children with birth defects [5, 7, 25, 34, 37]. Maternal characteristics were age at delivery, race/ethnicity, nativity, education, marital status, and rural residency. Infant characteristics were sex, pre-term birth (<37 weeks gestation), low birth weight (<2500 g), and postneonatal death during the study period. Health care system characteristics were adequacy of prenatal care, health insurance payer, inter-hospital transfer, and birth hospital nursery care level (I, II, or III) [3].

We identified adequacy of prenatal care using the Kotelchuck Index, which classifies prenatal care services based on the number and timing of prenatal care visits [16]. Due to small cell sizes, we reported adequacy of prenatal care as a binary variable, collapsing adequate and adequate plus care into a variable "adequate care," and intermediate and inadequate care into "less than adequate care."

We identified maternal residential rurality by comparing the geocoded maternal residential addresses reported at delivery with the 2000 US Census data that identified rurality at the block group level [39]. We created a dichotomous variable, "urban" or "rural" to describe residential rurality.

Health insurance payers were the expected payers for the birth hospitalization reported in the hospital discharge data. Payers for the birth hospitalization were defined as public (Medicare, Medicaid, and other state or local insurance, such as the Florida Children's Health Insurance Program, KidCare), private (private or employer-based insurance, including military coverage, such as Civilian Health and Medical Program of the Uniformed Services [CHAMPUS] or TriCare), or self-pay or under-insured (defined by the AHCA as no third party coverage or <30 % estimated insurance coverage) [12].

Inter-hospital transfers were identified when hospital discharge records showed that an infant was admitted to a hospital on the same day the infant was discharged from another hospital or if a 1-day difference existed between a discharge from one hospital and an admission to another hospital and the records included a "transfer" code [10]. Only inter-hospital transfers that occurred during the birth hospitalization were observed. We coded inter-hospital transfers as no transfer, transfer by day 3 of life, or transfer after day 3 of life.

Statistical analyses—We calculated the mean, median, and range in days for time-to-repair among the infants who had a surgical repair during the first year of life. In the bivariate analyses, we examined repair in ≤2 versus >2 days by maternal, infant, and health

care system characteristics. We used chi-square analyses for the categorical variables and Fisher's exact test for small cell sizes. p values <0.05 were considered statistically significant.

In multivariable analyses, we estimated unadjusted prevalence ratios (uPR), adjusted prevalence ratios (aPR), and corresponding 95 % confidence intervals (95 % CI), using modified log-linear Poisson regression with a robust variance estimate. We selected Poisson regression because it provides directly interpretable risk ratio results in analysis of dichotomous variables, especially when the outcome of interest is not rare [43].

Our multivariable models were based on the framework described by Aday and Andersen [1] and informed by previous research [5, 25, 34, 37]. We assessed for multicollinearity using the variance inflation factor and excluded a variable if it was closely correlated with another variable (e.g., hospital transfers were correlated with nursery care level) or if the category size was too small to support the analysis. We did not think death beyond 1 month of life would have bearing on the outcome, so did not control for postneonatal death. Our final models included the following variables: maternal age, race/ethnicity, education, and nativity; marital status; residential rurality; infant's sex; preterm birth; co-occurring hydrocephalus; SB type; adequacy of prenatal care; birth hospital nursery care level; and health care payer.

We conducted analyses using SAS 9.2 (SAS Institute, Inc., Cary, NC). The Institutional Review Boards at the University of North Carolina at Charlotte, the FDOH, and CDC approved this study.

Results

Study sample

Of 614 infants in the FBDR who had ICD-9-CM codes for SB without anencephaly, 569 were linked to a birth hospitalization in the hospital discharge records. Among all infants with a birth hospitalization (n = 569), 299 (52.5 %) had a recorded postnatal surgical repair and survived the neonatal period. Among the 299 infants, 215 (71.9 %) had a coded MMC repair. We excluded 16 infants whose time-to-repair was greater than 21 days [14] to ensure we captured the primary surgical repair for MMC. Our final analytic sample included 199 infants.

Descriptive and bivariate results

Table 1 summarizes selected maternal, infant, and health care system characteristics. About 52 % (n = 104) of mothers were non-Hispanic White, and 77.9 % (n = 155) of mothers were born in the USA. About 20 % of infants (n = 39) had non-isolated SB and 87.9 % (n = 175) had hydrocephalus. Approximately 25 % (n = 50) of infants were born preterm.

We found that 76.4 % (n = 152) of infants had surgical repair by day 2 (data not shown). About 14 % (n = 28) had a surgical repair between days 3 and 7; 9.5 % (n = 19) had a surgical repair after day 7. Mean time-to-repair for all infants was 2.5 days (standard deviation, 3.7 days) and median time-to-repair was 1.0 day (interquartile range, 1.0 day)

(results not shown). Although mean time-to-repair varied, infants with isolated SB, non-isolated SB, and hydrocephalus all had a median time-to-repair of 1.0 day (results not shown).

SB type was not associated with time-to-repair ($p = 0.452$). However, the infants with hydrocephalus ($p = 0.006$) and those born in a hospital with level III nursery care ($p = 0.003$) were more likely to have timely SB repair (Table 1).

Multivariable results

After multivariable adjustment, the presence of hydrocephalus and preterm birth were associated with an increased likelihood for timely repair (aPR = 1.48, 95 % CI 1.02–2.14 and (aPR = 1.19, 95 % CI 1.01–1.41, respectively). Infants who were born in a hospital with a lower nursery care level (I or II) were less likely to have a timely repair (aPR: 0.71, 95 % CI 0.52–0.98) than infants born in a hospital with level III nursery care (Table 2).

Discussion

Consistent with our first hypothesis, the majority of infants who had a postnatal surgical repair of SB had their repair within the first few days of life. The proportion of infants who had a repair at ≤2 days was somewhat lower (76.4 %) than reported in other recent studies [17, 33]. However, one study excluded roughly 40 % of hospital discharges for infants with surgical repair for SB that did not have either both a procedural code for MMC repair and a known age at repair or a code for MC repair during the first 4 days [17]. In contrast, our findings for a cohort of infants with a coded MMC repair provide the first population-based estimate of the frequency of the first surgical repair in infants with SB before and after 2 days.

Consistent with our second hypothesis, infants who had hydrocephalus were more likely to have a timely repair than infants who did not have hydrocephalus. Our finding of timely repair among almost 90 % of infants with hydrocephalus suggests that most infants with the most severe form of SB had a surgical repair within the first 2 days of life [21].

We found that infants born in a hospital with level I or II nursery care were less likely to have a timely surgical repair. The lower likelihood of timely repair among infants born in hospitals with lower levels of nursery care may result from lack of prenatal diagnoses and subsequent lack of appropriate referrals, the added time needed for transfer to a hospital with higher level of nursery care for treatment, or because of less medical need.

Preterm infants with SB also were more likely to have a repair of MMC ≤2 days compared to term infants. Although we hypothesized that this finding was associated with the birth hospital level of nursery care, we found no evidence of confounding or correlation between preterm birth and level of nursery care in relation to timing of repair.

Finally, we found no differences in the timing of SB repair by SB type. The additional diagnoses associated with non-isolated SB may not require care that would influence surgical repair of SB.

Limitations and strengths

This study was limited by several factors. The FBDR identifies infants using passive surveillance methodologies, which may lead to under-reporting or misreporting of infants with birth defects. However, the FBDR's overall case ascertainment for SB without anencephaly was 88.0 % [30, 31]. In addition, ICD-9-CM diagnostic codes do not differentiate between MMC, MC, and myelocele cases.

We unexpectedly found that only 52.5 % of infants who had a recorded birth hospitalization and survived the neonatal period had a recorded postnatal surgical repair. We examined nationwide claims data for both publicly insured and privately insured infants with spina bifida hospitalizations and found similarly low rates of documented surgical repair. Future research comparing surgical repairs recorded in medical records with repairs reported in administrative data is needed to explain what appear to be low rates of surgical repair of MMC in both the Florida data and other nationwide databases.

Although data from 108 Florida hospitals are represented in these data [11], not all hospitals are required to report to the AHCA. Most non-reporting hospitals do not provide newborn care [13]. However, the Shriners Hospital for Children in Tampa, Florida, is a non-reporting hospital that specializes in providing surgical care for children and maintains an out-patient SB clinic. We were unable to obtain data from the Shriners Hospital for Children in Tampa. A report on the economic costs of birth defects estimated that the two Shriners hospitals in California accounted for less than 2 % of medical costs associated with SB [41]; this result suggests that the number of repairs at the Shriners Hospital in Tampa may have been modest. Although the lack of data from the Shriners Hospital is a limitation, it should not bias our results.

We were also not able to determine if an infant had a pre-natal surgical repair of SB. While no information on prenatal surgical repair of SB was available from the data, we know that no prenatal repairs occurred in the study sample in Florida after February 2003 because of hospital agreements associated with the Management of Myelomeningocele Study (MOMS) clinical trial [2]. We had no information on prenatal repairs that may have occurred before that time, however. The effects of the concurrent MOMS research on the outcomes of our study are unknown.

Our study also has several strengths. This study used statewide, population-based birth defects registry data of unduplicated infants from a large, racially, and ethnically diverse population [15]. In addition, our data included hospitalizations for unique infants followed over time, rather than using data from unidentified hospitalizations, which can represent the same infants admitted multiple times. Finally, we included both publicly insured and privately insured infants in the analyses.

Conclusions

Results of this study showed that most infants with post-natal closure of SB had the repair in the first 2 days of life. Infants with hydrocephalus, a likely proxy for MMC, were more likely to have a timely repair. Infants born in hospitals with lower level nursery care were less likely to have a timely repair.

These findings have implications for public health. Programs that advocate for SB awareness, such as the Spina Bifida Association of Central Florida's 2012 campaign *Redefining Spina Bifida* [35], are important for increasing public awareness of prenatal screening for SB. Prenatal diagnosis of SB can facilitate counseling [29, 32] and planning for birth in a hospital with a higher level of nursery care, thus improving the opportunity for timely repair.

Timeliness of postnatal surgical repair of SB warrants further research on several fronts. Collaborative multi-state, population-based studies linking multiple birth defects registries and hospital discharge data would be useful to further examine timeliness of surgical repair of SB and related factors. An understanding of patterns and predictors of timely care is important to inform coordination of service delivery and adherence to care standards by health planners and practitioners, particularly those serving CSHCN, such as children with birth defects like SB.

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Table 1

Selected characteristics of Florida-born infants with spina bifida (SB) by timing of postnatal surgical repair, 1998–2007

Characteristics	All infants (*n* = 199)		Time-to-repair				*p* value
			Timely repair day 0–2 (*n* = 152)		Later repair after day 2 (*n* = 47)		
	n	(%)	*n*	(%)	*n*	(%)	
Maternal							
Age (years)							
<25	86	(42.3)	65	(42.8)	21	(44.7)	0.876
25–29	52	(26.1)	39	(25.7)	13	(27.7)	
≥30	61	(30.6)	48	(31.6)	13	(27.7)	
Race/ethnicity							
Non-Hispanic White	104	(52.3)	80	(52.6)	24	(51.1)	0.978
Non-Hispanic Black	43	(21.6)	33	(21.7)	10	(21.3)	
Hispanic	49	(24.6)	37	(24.3)	12	(25.5)	
Other	NR	NR	NR	NR	NR	NR	
Nativity							
Born in United States	155	(77.9)	117	(77.0)	38	(80.9)	0.576
Foreign-born	44	(22.1)	35	(23.0)	9	(19.1)	
Marital status							
Married	118	(59.3)	87	(57.2)	31	(66.0)	0.288
Not married	81	(40.7)	65	(42.8)	16	(34.0)	
Education							
No high school diploma	50	(25.4)	36	(24.0)	14	(29.8)	0.426
High school diploma or more	147	(74.6)	114	(76.0)	33	(70.2)	
Missing	NR	NR	NR	NR	NR	NR	
Residential rurality[a]							
Urban/urban cluster	165	(82.9)	127	(83.6)	38	(80.9)	0.667
Rural	34	(17.1)	25	(16.4)	9	(19.1)	
Infant							
Sex							
Female	92	(51.8)	74	(48.7)	18	(38.3)	0.212

Characteristics	All infants (*n* = 199)		Time-to-repair				*p* value
			Timely repair day 0–2 (*n* = 152)		Later repair after day 2 (*n* = 47)		
	n	(%)	*n*	(%)	*n*	(%)	
Male	107	(48.2)	78	(51.3)	29	(27.1)	
SB type[b]							
Isolated	160	(80.4)	124	(81.6)	36	(76.6)	0.452
Non-isolated	39	(19.6)	28	(18.4)	11	(23.4)	
Hydrocephalus							
No	24	(12.1)	13	(8.6)	11	(23.4)	*0.006*
Yes	175	(87.9)	139	(91.4)	36	(76.6)	
Preterm birth (<37 weeks gestation)							
No	149	(74.9)	109	(71.7)	40	(85.1)	0.064
Yes	50	(25.1)	43	(28.3)	7	(14.9)	
Low birth weight (<2500 g)							
No	172	(86.9)	131	(86.8)	41	(87.2)	0.932
Yes	26	(13.1)	20	(13.3)	6	(12.8)	
Death[c]							
No death	190	(95.5)	144	(94.7)	46	(97.9)	0.452
Death during infancy	NR	NR	NR	NR	NR	NR	
Death during ages 1–4 years	NR	NR	NR	NR	NR	NR	
Health care system							
Prenatal care[d]							
Adequate care	146	(73.3)	109	(73.2)	37	(74.0)	0.362
Less than adequate care	43	(21.6)	35	(23.4)	8	(16.0)	
Missing	10	(0.1)	NR	NR	NR	NR	
Payer at birth hospitalization[e]							
Public payer	111	(55.8)	83	(54.6)	28	(59.6)	0.549
Private payer	88	(44.2)	69	(45.4)	19	(40.6)	
Birth hospital nursery care level[f]							
Level III	167	(83.9)	134	(88.2)	33	(70.2)	*0.003*
Level I or II	32	(16.1)	18	(11.8)	14	(29.8)	

Characteristics	All infants (*n* = 199)		Time-to-repair				*p* value
			Timely repair day 0–2 (*n* = 152)		Later repair after day 2 (*n* = 47)		
	n	(%)	*n*	(%)	*n*	(%)	
Inter-hospital transfer[g]							
No inter-hospital transfer	137	(68.8)	110	(72.4)	27	(57.5)	0.116
Transfer within 3 days of birth	61	(30.6)	41	(27.0)	30	(42.6)	
Transfer after 3 days	NR	NR	NR	NR	NR	NR	

Columns may not add to 100 % because of missing or unknown values

p values compare repairs by day 2 to those after day 2 and are considered statistically significant at <0.05, noted in italics

NR not reported due to small cell counts

[a]Residential rurality was identified using geocoded maternal residence and 2000 US Census data

[b]Isolated SB was defined as SB with no additional major coded defects other than the sequence of defects related to SB

[c]All deaths occurred during study period, January 1, 1998, through December 31, 2008

[d]Adequacy of prenatal care was determined using the Kotelchuck Index. Based on Kotelchuck scoring, adequate and adequate plus were considered “adequate care”; inadequate and intermediate care were considered “less than adequate care”

[e]Public insurance included Medicare, Medicaid, and KidCare insurance. Private included employer-based insurance, including military coverage [Civilian Health and Medical Program of the Uniformed Services (CHAMPUS) or TriCare]

[f]Level III is the highest level of hospital nursery care

[g]Inter-hospital transfers were identified when hospital discharge records showed that an infant was admitted to a hospital on the same day the infant was discharged from another hospital or if a 1-day difference existed between a discharge from one hospital and an admission to another hospital

Table 2

Unadjusted (uPR) and adjusted (aPR) modified Poisson regression results for the association between selected characteristics and time-to-repair among Florida-born infants with spina bifida (SB), 1998–2007 (n = 199) [≤2 vs. >2 days (reference)]

Characteristics	All infants with SB (n = 199)			
	Unadjusted		Adjusted	
	uPR	(95 % CI)	aPR	(95 % CI)
Maternal				
Age (years)				
<25	0.97	(0.82–1.14)	0.94	(0.77–1.15)
25–29	1.00		1.00	
≥30	1.15	(0.87–1.22)	0.95	(0.82–1.21)
Race/ethnicity				
Non-Hispanic White	1.00		1.00	
Non-Hispanic Black	1.02	(0.84–1.24)	0.96	(0.77–1.20)
Hispanic	0.97	(0.79–1.18)	0.99	(0.79–1.25)
Nativity				
Born in United States	1.00		1.00	
Foreign-born	1.04	(0.87–1.25)	1.12	(0.93–1.35)
Marital status				
Married	1.00		1.00	
Not married	1.11	(0.95–1.30)	1.14	(0.94–1.40)
Education				
High school diploma or more	1.00		1.00	
No high school diploma	0.90	(0.73–1.11)	0.88	(0.71–1.09)
Residential rurality[a]				
Urban/urban cluster	1.00		1.00	
Rural	0.93	(0.74–1.18)	0.96	(0.76–1.22)
Infant				
Sex				
Female	1.00		1.00	
Male	0.89	(0.76–1.04)	0.91	(0.78–1.07)
SB type[b]				
Isolated	1.00		1.00	
Non-isolated	0.99	(0.81–1.22)	1.05	(0.86–1.29)
Hydrocephalus				
No	1.00		1.00	
Yes	*1.46*	(*1.01–1.37*)	*1.48*	(*1.02–2.14*)
Preterm birth (< 37 weeks)				
No	1.00		1.00	
Yes	*1.17*	(*1.01–1.37*)	*1.19*	(*1.01–1.41*)
Health care system				

Characteristics	All infants with SB (*n* = 199)			
	Unadjusted		Adjusted	
	uPR	(95 % CI)	aPR	(95 % CI)
Prenatal care[c]				
Adequate care	1.00		1.00	
Less than adequate care	1.08	(0.91–1.29)	1.11	(0.93–1.33)
Payer at birth hospitalization[d]				
Private payer	1.00		1.00	
Public payer	0.96	(0.82–1.12)	0.88	(0.74–1.05)
Birth hospital nursery care level[e]				
Level III	1.00		1.00	
Level I or II	0.72	(0.52–1.00)	*0.71*	(*0.52–0.98*)

Values in italics are statistically significant. Adjusted model was adjusted for all covariates in table

uPR unadjusted prevalence ratio, *aPR* adjusted prevalence ratio, *95 % CI* 95 % confidence interval

[a] Residential rurality was identified using geocoded maternal residence and 2000 US Census data

[b] SB type: isolated SB was SB with no additional coded major defects other than the sequence of defects related to SB

[c] Adequacy of prenatal care was determined using the Kotelchuck Index. Based on Kotelchuck scoring, adequate and adequate plus were considered "adequate care"; inadequate and intermediate care were considered "less than adequate care"

[d] All payers were expected payers. Public insurance included Medicare, Medicaid, and KidCare insurance. Private included employer-based insurance, including military coverage [Civilian Health and Medical Program of the Uniformed Services (CHAMPUS) or TriCare]

[e] Level III is the highest level of hospital nursery care