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Pregnancy among mothers with spina bifida

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Summary

Introduction
Recognizing the importance of sexual and reproductive health to patients with spina bifida (SB), pediatric urologists have taken responsibility for initiating conversations regarding this topic with adolescent and young adult SB patients. However, the sexual and reproductive health of women with SB remains under-investigated. It is unknown how many women are having babies, what mode of delivery is used, and if this has changed over time with the increasing life expectancy of these patients. A better understanding of pregnancy and delivery among young women with SB will enable urologists to provide more informed, comprehensive counseling to patients.

Objective
We sought to compare hospitalizations for delivery in women with and without SB to determine differences in the mode of delivery used and changes in the rate of deliveries over time.

Study design
Using the Healthcare Cost and Utilization's National Inpatient Sample, we identified all hospitalizations for delivery in 2003–2013. After distinguishing between hospitalizations among women with and without SB, temporal trends analysis and bivariate comparison were performed to determine differences in patient and hospital characteristics and mode of deliveries.

Results
We identified 10,147 hospitalizations for deliveries among women with SB and 42,197,763 among women without. Of all hospitalizations for deliveries, the percentage of deliveries by women with SB increased by 56% between 2003 and 2013 (629–925 deliveries per year, $p < 0.001$). Women with SB hospitalized for a delivery differed from those without SB. They had a higher number of comorbidities and were more likely to be white, have Medicare or private insurance, live outside a city, and deliver at an urban teaching hospital (all $p < 0.001$). Women with SB were significantly more likely to undergo a caesarean section (see Figure, 52.4% of women with SB vs. 31.9% of those without, $p < 0.001$), although nearly half were able to undergo vaginal delivery. For women with SB, 25.9% of all deliveries occurred by age 22, which did not differ significantly from women without SB (24.7% of all deliveries).

Discussion
There are significant differences in the characteristics and mode of delivery between women with and without SB who are hospitalized for a delivery. The number of deliveries among these women are significantly increasing and over a quarter of the deliveries occur by age 22.

Conclusion
With increasing rates of deliveries and young age at delivery for women with SB, it is imperative that pediatric and transitional urologists initiate discussions on sexual and reproductive health beginning in adolescence.

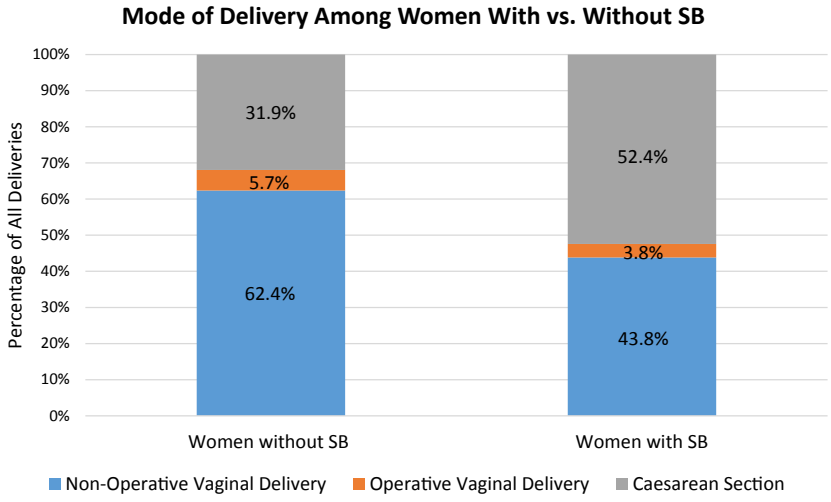

Figure Mode of delivery used by women with and without spina bifida in 2003–2013.

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Introduction

Historically, only 60% of infants born with spina bifida (SB) survived beyond the first year of life [1], but the past few decades have been transformative for people born with this birth defect. Innovations such as ventricular shunts for treatment of hydrocephalus and urinary diversion procedures for preserving renal function have greatly improved survival such that, for the first time, approximately 85% of people born with SB are now surviving into adulthood [2] As a consequence, the providers who care for these patients are managing health issues that were previously not a concern.

Perhaps no health issue has been more under-investigated in the SB population than female sexual and reproductive health. Yet this topic is important to quality of life for these women, many of whom would like to have children of their own [3–5]. As genitourinary specialists with expertise in SB care, urologists have recognized our role in discussing sexual and reproductive health with our SB patients [6,7]. Therefore, a better understanding of pregnancy and delivery among women with SB is needed to enable urologists to provide more comprehensive and informed counseling to these women. Currently it is unknown how many women with SB of child-bearing age are having children, or if this number has been increasing with improved survival of SB women. It is also unknown what modes of delivery are being used for these women and if these differ from women without SB.

In this context, we conducted a retrospective cohort study using national discharge data. After identifying hospitalizations for a delivery among pregnant women with and without SB, we determined who is having children, where, trends over time, and how these two groups of women compare. We hypothesized that as more patients with SB are living into adulthood, more are also becoming mothers. Additionally, we hypothesized that caesarean section is the dominant mode of delivery for women with SB. Findings from our study have clinical implications for urologists who, given their prominent role in SB management across a life course, are well-positioned to counsel patients on this topic.

Materials and methods

Data source and study population

For this study, we analyzed data from the National (Nationwide) Inpatient Sample (NIS), which was developed by the Healthcare Cost and Utilization Project and sponsored by the Agency for Healthcare Research and Quality. The NIS is the largest all-payer inpatient database in the USA; it is a 20% sample of discharges from participating hospitals, weighted estimates from which are generalizable to the universe of hospital discharges [8]. Prior to 2012, the NIS was a sample of discharges from all participating hospitals; weighted estimates are used to allow for comparisons before and after this revision. The sample frame includes all women hospitalized for a delivery between 2003 and 2013.

First, all women discharged from the hospital after any type of delivery were classified through Clinical Classification Software groups (Appendix 1). We then distinguished between deliveries among women with and without SB using relevant International Classification of Diseases, Ninth Revision (ICD-9), Clinical Modification diagnosis codes (Appendix Table 1). We also distinguished among women with SB occulta, SB without hydrocephalus, and SB with hydrocephalus (Appendix Table 1).

Statistical analysis

For our initial analytic step, we examined differences between women with and without SB who were hospitalized for delivery. Specifically, the women were compared with respect to a variety of sociodemographic factors including their age on admission, race/ethnicity (white, black, Hispanic, or other), income (based on ZIP-code level data), insurance status (self-pay, private, Medicaid, Medicare, no charge, or other), and level of urbanicity. The patients' level of comorbidity was also compared using the Elixhauser method [9]. The hospitals where these women were admitted were characterized based on their bed size, teaching status, and ownership. For these bivariate comparisons, parametric and non-parametric tests were used where appropriate.

We then measured rate of hospitalization for delivery (per 100,000) among women with and without SB and among the subtypes of SB. The frequency of the various modes of delivery (non-operative vaginal, operative vaginal, or cesarean) within each group was calculated. Operative vaginal delivery was defined as a delivery which requires vacuum extraction, the use of forceps, or breech delivery.

Finally, we fit multinomial logistic regression models to evaluate significant changes over time in rates of delivery and mode of delivery among women with and without SB. All analyses were performed at the type I error level of 0.05 using SAS version 9.4 (Cary, NC, USA).

Results

We identified 10,147 hospitalizations for deliveries among women with SB and 42,197,763 among women without. Of the women with SB, 47.7% did not have hydrocephalus, 18.7% had hydrocephalus, and 33.6% had spina bifida occulta. Women with SB hospitalized for a delivery differed from those without SB in several significant ways. They had a higher mean number of comorbid conditions (0.66 vs. 0.33 in women without SB, $p < 0.001$). They were also more likely to be white, have private insurance or Medicare, live outside a city, and deliver at an urban teaching hospital (all $p < 0.001$) (Table 1).

Of all hospitalizations for deliveries, the percent of deliveries by women with SB increased by 56% between 2003 and 2013 (629–925 deliveries per year, $p < 0.001$; Fig. 1). The mean age at delivery for women with SB was 27.3 years (standard deviation (SD) 12.8); the mean age at delivery for women without SB was 27.6 years (SD 13.4). Of all hospitalizations for deliveries in each group, 4.6% occurred before age 18 in women with SB versus 5.4% for women without ($p = 0.426$). Approximately a quarter of hospitalizations for delivery occurred by age 22 in each group (25.9% for women with SB and 24.7% for women without). However, a

Table 1 Personal and hospital characteristics of women hospitalized for a delivery.

	Healthy females $N = 42,197,763$	Females with spina bifida $N = 10,147$	p-value
Patient characteristics			
Age, years			0.0023
<18	2,277,942 (5.4)	469 (4.6)	
18–22	8,164,053 (19.3)	2159 (21.3)	
23–26	7,841,041 (18.6)	2015 (19.9)	
27–30	9,737,067 (23.1)	2430 (24.0)	
31–34	8,026,953 (19.0)	1855 (18.3)	
>34	6,150,707 (14.6)	1219 (12.0)	
Race/ethnicity			<0.001
White	17,942,976 (42.5)	6056 (59.7)	
Black	4,614,903 (10.9)	626 (6.2)	
Hispanic	8,031,276 (19.0)	1110 (10.9)	
Asian or Pacific Islander	1,733,209 (4.1)	157 (1.5)	
Native American	252,348 (0.6)	38 (0.4)	
Other	1,595,831 (3.8)	350 (3.4)	
Unknown	8,027,218 (19.0)	1810 (17.8)	
Mean number of Elixhauser conditions	0.33	0.66	<0.001
Median household income			0.022
Very low	11,488,543 (27.2)	2458 (24.2)	
Low	10,616,708 (25.2)	2705 (26.7)	
Medium	10,367,717 (24.6)	2624 (25.9)	
High	9,724,793 (23.0)	2358 (23.2)	
Insurance			<0.001
Self-pay	1,241,607 (2.9)	143 (1.4)	
Private	21,715,775 (51.5)	5399 (53.2)	
Medicare	244,731 (0.6)	392 (3.9)	
Medicaid	17,802,582 (42.2)	3867 (38.1)	
No charge/other	1,193,067 (2.8)	346 (3.4)	
Residence			<0.001
Central/Fringe (1 million or greater)	24,187,113 (57.3)	5274 (52.0)	
50,000–999,999	11,844,581 (28.1)	3115 (30.7)	
10,000–49,999	3,839,656 (9.1)	1142 (11.3)	
<10,000	2,326,412 (5.5)	616 (6.1)	
Hospital characteristics			
Bed size			0.005
Small	4,699,098 (11.1)	897 (8.8)	
Medium	11,193,833 (26.5)	2645 (26.1)	
Large	26,304,830 (62.3)	6605 (65.1)	
Location and teaching status			<0.001
Rural	4,781,284 (11.3)	1116 (11.0)	
Urban non-teaching	17,957,534 (42.6)	3700 (36.5)	
Urban teaching	19,458,944 (46.1)	5331 (52.5)	

significantly greater percentage of all hospitalizations for delivery occurred after age 34 for women without SB, compared with those with SB (14.6% vs. 12%, $p = 0.007$).

The mode of delivery differed significantly between women with and without SB ($p < 0.001$). Although 43.8% of women with SB were able to have a normal vaginal delivery, this was less than the 62.4% of women without SB. Conversely, women with SB were significantly more likely to undergo cesarean section (52.4% of women with SB compared with 31.9% of those without). The mode of delivery also differed significantly among women with SB by severity (Table 2). Women with SB occulta were most likely to have a normal vaginal delivery (54.8%), whereas women with SB and hydrocephalus were most likely to undergo a cesarean section (64.3%, $p < 0.001$). Although the total number of deliveries among women with SB increased between 2003 and 2013, there has been no significant change in the mode of delivery for these women over this time period (Fig. 2). However, there was a slight increase in the rates of cesarean section and decrease in the rates of operative vaginal deliveries for women without SB over the same time period (Fig. 3; $p < 0.001$).

Discussion

Rates of hospitalization for a delivery among women with SB are significantly increasing. Just over half of these

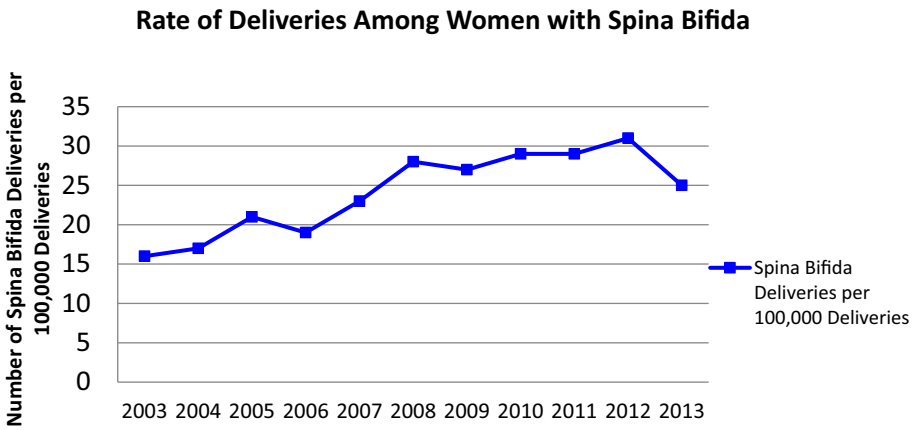

Figure 1 Temporal trend of rate of hospitalization for delivery among women with spina bifida per 100,000 deliveries.

Table 2 Delivery type by spina bifida classification.

	Spina bifida type		
Mode of delivery	Occulta	Without hydrocephalus	With hydrocephalus
Normal vaginal delivery	1869 (54.8)	1974 (40.8)	603 (31.9)
Assisted vaginal delivery	125 (3.7)	185 (3.8)	72 (3.8)
C-section	1416 (41.5)	2685 (55.4)	1218 (64.3)

women undergo a cesarean section, while the remainder are able to undergo a vaginal delivery. Conversely, only 32% of women without SB underwent a cesarean section. This trend remained consistent over time for SB women. Over a quarter of the deliveries occur by age 22, which is typically considered the "transitional period" between pediatric and adult clinics for these patients.

Current knowledge about pregnancy in SB has been limited to small case series [10—12]. It was formerly preferred for women with SB to undergo a caesarean section [13]; however, increasingly there are reports of the safety of vaginal deliveries among carefully selected women with SB [10,14,15]. Many experts now recommend vaginal delivery unless there are obstetrical reasons for cesarean section such as a narrow pelvis, significant scoliosis, significant contractures, or pelvic organ prolapsed [4,12,15]. Additionally, difficulty with Valsalva during labor could be an indication for cesarean section [4,15], although the uterus has been shown to contract spontaneously after the nerve supply has been severed, allowing for successful vaginal delivery in 21 women with paraplegia in one study [16]. Previous augmentation cystoplasty is not thought to be a contraindication for vaginal delivery [4,10,14] Whether cesarean section is indicated after bladder neck reconstruction or artificial urinary sphincter (AUS) placement remains uncertain. Thomas and Adams recommend cesarean section in patients who have undergone bladder neck reconstruction because of recurrent incontinence reported after vaginal delivery in non-neurogenic women who had undergone bladder neck

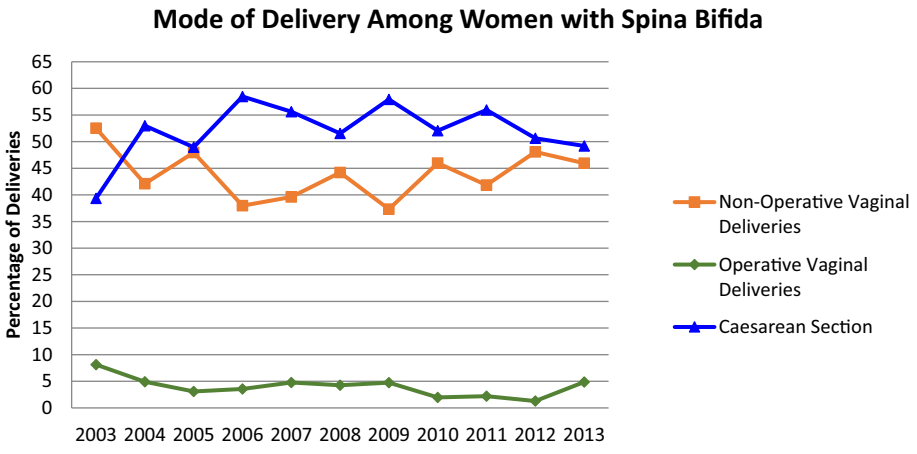

Figure 2 Temporal trends of the mode of delivery among women with spina bifida.

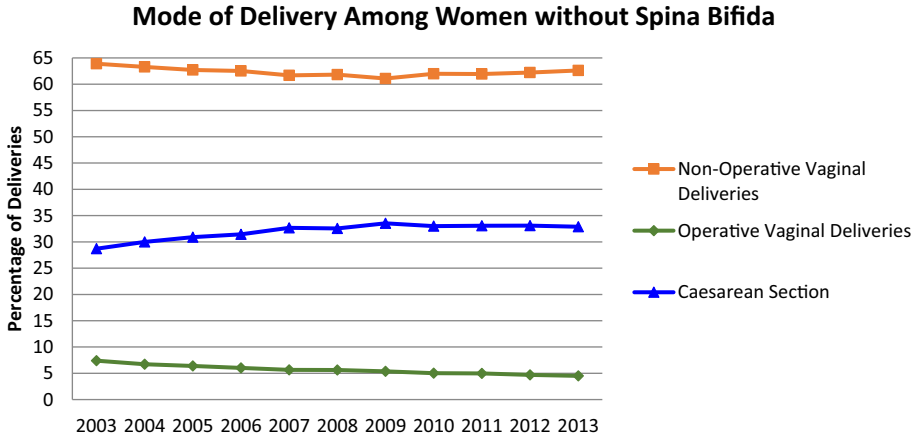

Figure 3 Temporal trends of the mode of delivery among women without spina bifida.

slings [4]. The best mode of delivery after AUS is less clear because of a paucity of data. Creagh et al. described successful vaginal deliveries in 11 patients who had undergone an AUS with return to continence in all patients by 4 weeks after delivery [14]. For women with an AUS who do undergo a vaginal delivery, Thomas and Adams suggest avoiding the use of forceps [4]. A midline uterine incision as opposed to a low transverse incision is recommended for cesarean section for previously augmented patients to avoid injury to the vascular pedicle [15]. A reconstructive surgeon should be present to help manage the reconstructed bladder intraoperatively [4,15].

There are limitations to using a national sample. The analysis relies on the accuracy of coding on the discharge record. While this limitation could result in an underestimation of the number of women with SB who were hospitalized for a delivery, ICD-9 coding has been demonstrated to be reliable for the identification of adults with congenital disease [17]. However, the accuracy of using ICD-9 codes to distinguish between the types of SB is less clear. Additionally, this study only includes women hospitalized for a delivery and therefore does not include women who underwent a birth outside of a hospital. Finally, ICD-9 coding currently does not stratify based on the severity of SB, so further details about these women with SB such as ambulatory status remain unknown. However, the use of a national sample as opposed to individual institutional reviews allows for a better understanding of trends for a relatively rare event among a population with a rare congenital defect.

Over a quarter of the deliveries occurred by age 22, which is the typical time of transition for these patients. Thus it is imperative that pediatric and transitional urologists discuss sexual and reproductive health with young women with SB. Leading experts in SB care have recognized that urologists need to take the lead in these discussions [6,7]; nonetheless, as few as 5% of young patients with SB have ever discussed sexual health with a physician [18]. There are many reasons why these discussions are so important. Sexual health is important to quality of life for these patients and many patients desire to discuss this topic with their physicians [3,4,19]. Many patients are also interested in having a family, and should understand the implications of pregnancy and delivery specific to spina bifida [3,5]. Additionally, many patients have misconceptions about their sexual health that should be addressed, such as falsely assuming they cannot get STDs [3,5,20]. Finally, we need to serve as the bridge to gynecologic care so these patients can get the screening as appropriate and have access to family planning.

Interestingly, recent research by Ebert et al. demonstrated that among a cohort of 21 female patients with exstrophy-epispadias with a mean age of 26, 62% were sexually active on a regular basis and became sexually active at a mean age of 17 [21]. Three females had delivered children at a mean age of 22 at the first pregnancy. Another three females were planning on becoming pregnant. These patients have many important similarities to patients with SB. Their young age at first intercourse and pregnancy further prove the importance of pediatric and transitional urologists providing sexual and reproductive education to urologic congenital patients.

As deliveries among women with SB are significantly increasing and many are occurring at a young age, it is time for us to start talking about sex with our patients. Exploratory research is needed to learn what to discuss, how, and when in a manner specific to young women with SB.

Conclusion

The number of deliveries among women with SB is significantly increasing and a slight majority of these women undergo a cesarean section. Urologists should be knowledgeable about the reproductive health of women with SB to offer counseling for these patients.

Conflict of interest

None.

Funding

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Appendix 1. ICD-9-CM codes associated with each clinical classification software group

A 194: Forceps delivery.

ICD-9-CM codes: 66950 66951

A 196: Other pregnancy and delivery including normal.

ICD-9-CM codes: 650 65100 65101 65110 65111 65120 65121 65170 65171 65173 65180 65181 65190 65191 V220 V221 V222 V240 V241 V242 V270 V724 V7242 V9100 V9101 V9102 V9103 V9109 V9110 V9111 V9112 V9119 V9120 V9121 V9122 V9129 V9190 V9191 V9192 V9199

B134: Cesarean section.

ICD-9-CM codes: 740 741 742 744 7499

B 135: Forceps; vacuum; and breech delivery.

ICD-9-CM codes: 720 721 7221 7229 7231 7239 724 7251 7252 7253 7254 726 7271 7279 728 729 733

B136: Artificial rupture of membranes to assist delivery.

ICD-9-CM codes: 7301 7309

B137: Other procedures to assist delivery.

ICD-9-CM codes: 731 7321 7322 734 7351 7359 738 7391 7392 7393 7394 7399

Appendix Table 1 CCS and ICD-9-CM codes included.

Diagnosis	Codes included
Non-operative vaginal delivery	CCS groups: A 196 (all normal deliveries including multiple gestations) B 136 (artificial rupture of membranes to assist delivery) B 137 (other procedures to assist delivery)
Operative vaginal delivery	CCS groups: A 194 (forceps delivery) B 135 (forceps, vacuum, and breech delivery)
Cesarean section	CCS groups: B 134 (cesarean section) ICD-9-CM code: 669.7
Spina bifida	ICD-9-CM codes: 741.00, 741.01, 471.02, 741.03 (SB with hydrocephalus) 741.90, 741.91, 741.92, 741.93 (SB without hydrocephalus) 756.17 (SB occulta)

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