Published in final edited form as:

J Pediatr Rehabil Med. 2017 December 11; 10(3-4): 303–312. doi:10.3233/PRM-170460.

Variation in surgical management of neurogenic bowel among centers participating in National Spina Bifida Patient Registry

Jonathan C. Routh[a,*], **David B. Joseph**[b], **Tiebin Liu**[c], **Michael S. Schechter**[d], **Judy K. Thibadeau**[c], **M. Chad Wallis**[e], **Elisabeth A. Ward**[c,f], and **John S. Wiener**[a]

[a]Division of Urology, Duke University Medical Center, Durham, NC, USA

[b]Department of Urology, University of Alabama-Birmingham, Birmingham, AL, USA

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

[d]Division of Pediatric Pulmonary Medicine, Children's Hospital of Richmond at Virginia Commonwealth University, Richmond, VA, USA

[e]Division of Urology, Primary Children's Hospital, Salt Lake City, UT, USA

[f]Carter Consulting, Inc., Atlanta, GA, USA

Abstract

PURPOSE—Optimal management of neurogenic bowel in patients with spina bifida (SB) remains controversial. Surgical interventions may be utilized to treat constipation and provide fecal continence, but their use may vary among SB treatment centers.

METHODS—We queried the National Spina Bifida Patient Registry (NSBPR) to identify patients who underwent surgical interventions for neurogenic bowel. We abstracted demographic characteristics, SB type, functional level, concurrent bladder surgery, mobility, and NSBPR clinics to determine whether any of these factors were associated with interventions for management of neurogenic bowel. Multivariable logistic regression with adjustment for selection bias was performed.

RESULTS—We identified 5,528 patients with SB enrolled in the 2009–14 NSBPR. Of these, 1,088 (19.7%) underwent procedures for neurogenic bowel, including 957 (17.3%) ACE/cecostomy tube and 155 (2.8%) ileostomy/colostomy patients. Procedures were more likely in patients who were older, white, non-ambulatory, with higher-level lesion, with myelomeningocele lesion, with private health insurance (all $p < 0.001$), and female ($p = 0.006$). On multivariable analysis, NSBPR clinic, older age (both $p < 0.001$), race ($p = 0.002$), mobility status ($p = 0.011$), higher lesion level ($p < 0.001$), private insurance ($p = 0.002$) and female sex ($p = 0.015$) were associated with increased odds of surgery.

[*]Corresponding author: Jonathan C. Routh, Duke University Medical Center, DUMC Box 3831, Durham, NC 27710, USA. Tel. +1 919 684 6994; Fax: +1 919 681 5507; jon.routh@duke.edu.

Conflict of interest

The authors have no conflict of interest to report.

CONCLUSIONS—There is significant variation in rates of procedures to manage neurogenic bowel among NSBPR clinics. In addition to SB-related factors such as mobility status and lesion type/level, non-SB-related factors such as patient age, sex, race and treating center are also associated with the likelihood of undergoing neurogenic bowel intervention.

Keywords

Pediatrics; spina bifida; neurogenic bowel; clinical care variation

1. Introduction

Spina bifida (SB) without anencephaly is the most common non-chromosomal central nervous system defect, occurring in approximately 3/10,000 live births [1, 2]. Neurogenic bowel dysfunction is the norm in patients with SB and negatively impacts quality of life [3, 4]. Many individuals with SB undergo procedures to manage neurogenic bowel, such as creation of a continent stoma, an incontinent colostomy, or percutaneous cecostomy tube (Chait) placement.

In previous analyses, our group has noted significant variation in the use of urological surgeries for management of neurogenic bladder in individuals with SB [5]. A recent analysis of national data revealed that privately insured patients were more likely to undergo continent bladder augmentation instead of incontinent urinary diversion [6] The underlying clinical reasons behind this variation are not clear. To the best of our knowledge, no studies have yet examined whether similar variation exists for neurogenic bowel procedures.

The primary objective of this study is, therefore, to describe current patterns of care among NSBPR clinics. We hypothesized that significant variation exists among NSBPR clinics in the use of neurogenic bowel procedures. Our exploratory, secondary aim was to identify factors that may influence such variation.

2. Patients and methods

2.1. Data source

The NSBPR was established by the Centers for Disease Control and Prevention to support efforts directed toward improving the consistency and quality of care of SB patients and to provide an infrastructure to support SB clinical research. In 2009, the NSBPR began accruing patients at 10 clinics and expanded in 2011 to 21 other leading SB centers which could successfully participate in the registry. As of December 2014, the NSBPR had enrolled 5,596 patients with SB.

After institutional review board approval, participating clinics collected longitudinal data on individuals with SB [7,8]. Limited data were also collected on patients who were eligible but not enrolled (ENE) in the NSBPR. Clinics with small enrollment (< 30 patients) were excluded, leaving a total of 5,528 patients from 19 clinics for this analysis. At the initial visit, basic demographic/diagnostic information in addition to previous surgical procedures were collected from each consenting/assenting patient. At the initial visit and each

subsequent annual visit, information on insurance status, education, and employment were collected in addition to any interval surgeries, procedures, treatments, and outcomes.

2.2. Statistical analysis

Predictor variables were selected *a priori* based on biologic plausibility and/or demonstrated associations in the literature. Covariates included basic patient demographics and clinical variables captured in the NSBPR: age, gender, race/ethnicity (non-Hispanic white, non-Hispanic black, Hispanic or Latino, or other/refused/unknown), insurance payer (any private vs. non-private), spina bifida type (myelomeningocele (MMC) vs. non-MMC), functional level of SB lesion (thoracic, lumbar, or sacral), mobility status (community ambulator, household ambulator, therapeutic ambulator, non-ambulator, or not applicable due to age if < 2 years), treating SB center and whether patients had previously/concurrently undergone bladder reconstruction.

We performed bivariate tests of association between these predictor variables and our primary outcome of interest, i.e. whether a particular patient underwent a continent (appendicocecostomy/antegrade continence enema (ACE) stoma or cecostomy tube) or incontinent neurogenic bowel (ileostomy or colostomy) intervention. Two multivariable logistic regression models were created to adjust for confounding (covariates listed above); first, to predict whether or not an intervention was performed, and, if yes, then second to predict what type of intervention was performed (continent vs. incontinent).

A two-sided alpha of 0.05 and 95% confidence intervals (CI) excluding 1 were used as criteria for statistical significance. All analyses were performed using SAS Version 9.3 and R.

2.3. Selection bias analysis

Because of concerns that enrollment of patients into the NBSPR by SB centers was not random [9], we used previously described statistical methods in an attempt to control for selection bias in our analysis [10,11]. We first performed logistic regression among eligible and enrolled (EAE) patients to determine the association between various characteristics (age, gender, race/ethnicity, type of spina bifida diagnosis, functional level of lesion, and type of health insurance) and the odds of having the procedures of interest performed in that population; due to small numbers of predicted events at some centers, center was not included as a covariate in this model. Using beta coefficients from this model, we estimated predicted probability of having bowel surgery for each eligible and not enrolled (ENE) patient according to the known variables. Then, we used this probability to assign a surgery status (yes/no) to each of the 498 ENE patients using a Bernoulli (or binomial) trial. In this trial, a surgery status is generated at random for each patient with a probability of ‘success’ (surgery = yes) that is equal to the previously estimated probability of surgery. This simulation was executed 10,000 times for ENE patients; each execution was identified with a unique seed number. Once all ENE patients had been probabilistically assigned a surgery status, the datasets from ENE and enrolled patients were combined into one dataset for the probabilistic selection bias analysis. In this combined dataset, enrollment was included in the model.

Selection bias is present if surgery status differed by enrollment in the different strata of the predictor variables. To test for this, we separated the datasets by surgery status and used logistic regression to model the likelihood of enrollment for individuals in each dataset, adjusted for other characteristics shown to be associated with surgery in our previous models. From these logistic regression models, we then calculated anti-logs by exponentiating the beta-coefficients and calculated a ratio of selection probability ratios (RSPR) by dividing the anti-log obtained in the surgery dataset by the anti-log obtained in the non-surgical dataset for each stratum. Our final adjusted odds ratio (AOR) was then calculated by dividing observed OR by RSPR.

3. Results

3.1. Cohort demographics

In total, we included 5,528 patients with SB (Table 1). The median age of enrolled patients was 11.7 years (mean 13.4, range 0.1–82 years). A majority of patients was female (52.7%), non-Hispanic white (64.1%) with non-private insurance (52.9%). Most patients had MMC (79.5%) with a lumbar level lesion (53.5%). The majority of patients were community ambulators (53.6%).

3.2. Neurogenic bowel procedures in NSBPR ratients

Among NSBPR patients, 1,088 (19.7%) underwent 1,305 procedures for management of neurogenic bowel. These include 957 (17.3%) ACE or cecostomy tube placements and 155 (2.8%) ileostomy or colostomy surgeries. Some patients underwent multiple procedures.

3.3. Variation in neurogenic bowel procedures

On bivariate analysis (Table 2), patients who underwent neurogenic bowel procedures were more likely to be older, white, privately insured, non-ambulatory, and to have MMC and a higher lesion level (all $p < 0.001$) than patients who did not undergo surgery; females were also more likely to have procedures ($p = 0.006$). NSPBR clinic was also highly associated with whether or not a child underwent a neurogenic bowel procedure ($p < 0.001$).

On multivariable analysis (Table 3), NSBPR clinic, higher lesion level, older age (all $p < 0.001$), MMC ($p = 0.012$), non-Hispanic white race/ethnicity ($p = 0.002$), reduced mobility status ($p = 0.011$), private insurance ($p = 0.002$) and female gender ($p = 0.015$) were associated with neurogenic bowel procedures.

We then separately modeled (Table 4) variation in the use of incontinent diversions (ileostomy or colostomy) compared to continent surgeries (ACE or cecostomy tube). Patients who were non-Hispanic black had higher odds of having undergone ileostomy or colostomy surgery compared with non-Hispanic white patients ($p = 0.004$), as did younger patients, non-ambulatory patients, those who had not undergone bladder reconstruction, who had non-MMC lesions, and who had been treated at certain NSBPR clinics (all $p < 0.001$).

3.4. Selection bias

After adjusting for the likelihood of being enrolled if a patient had surgery (Table 5), the adjusted odds ratios did not significantly differ from the original odds ratios calculated solely based on enrolled patient data. Thus, there was little evidence of clinically significant selection bias among NSBPR centers, with no statistically significant covariates becoming insignificant or vice versa.

4. Discussion

In this national, multicenter study, we demonstrate an association between neurogenic bowel surgery and SB-related factors such as bladder surgery, mobility status and lesion type/level. In addition, non-SB-related factors such as patient age, gender, race and treating center were also associated with the likelihood of undergoing neurogenic bowel procedures.

This last association suggests the presence of significant variation in the use of procedures to manage neurogenic bowel among NSBPR clinics. The overall intervention rate across all clinics was 19%. At some clinics, however, only 3% of patients underwent neurogenic bowel procedures; at other clinics, up to 38% of patients did. This variation remained significant even after correcting for clinical and non-clinical factors.

Surgical management of neurogenic bowel has evolved significantly over the past several decades. Historically, incontinent diversions such as colostomy or ileostomy were the only means available to those who failed non-operative measures. In 1990, Malone et al. reported initial results with appendicocecostomy, also known as the ACE procedure, to allow antegrade enemas [12]. This procedure has been reported to have continence rates of 90% or better with low complication rates [12–15]. For patients without an appendix, who are not good surgical candidates, or who do not desire intraabdominal surgery, a cecostomy tube can be placed percutaneously to also allow antegrade enemas to achieve colonic evacuation. Among patients enrolled in the NSBPR, continent ACE or cecostomy tube procedures are performed $>$ 6-fold as commonly as incontinent diversions. This is reflective of national trends towards increased usage of continent rather than incontinent procedures among SB patients [6,13]. Perhaps unsurprisingly, bladder surgery (concurrent or prior) was associated with continent bowel procedures being performed. The drop-off in the frequency of these procedures in the oldest group of patients is likely explained by the fact that these continent procedures did not exist when these patients were younger. Likewise, adult patients may not have desired surgical interventions.

In a previous analysis, Sawin and colleagues analyzed continence rates among the first 10 clinics to participate in the NSBPR; they found that only 30% of SB patients reported themselves as being continent of stool [7]. However, this analysis did not examine whether patients undergoing ACE or colostomy procedures were more or less likely to identify themselves as “continent” compared to patients receiving medical management alone or no management. Given recent advances with cone enemas or the Peristeen® anal irrigation system, this is clearly an area that merits further study [16]. In terms of establishment of applicable standards of care, future analysis will need to focus on the outcomes of all interventions, specifically whether specific interventions or specific clinics with higher

surgical rates have higher continence rates. The NSBPR has been recently modified to allow better quantification and qualification of fecal incontinence episodes, so more granular data may be available in coming years.

In our analysis, we found that patients undergoing ACE or cecostomy tube procedures were more likely to be younger (predominantly aged 10–21 years), to be white, to be non-ambulatory, to undergo concurrent bladder reconstruction, and to have been treated at certain NSBPR clinics. Compared to incontinent procedures and to medically managed constipation, these procedures have been reported to significantly increase satisfaction and quality of life measures among SB patients [17,18]. Therefore, it is not surprising ACE/cecostomy procedures were much more frequently performed than incontinent procedures. Similarly, fecal stream diversion procedures such as colostomy may be performed as a component of management of posterior pelvis pressure ulcers that may be more common in adults who are heavier or possibly ambulate less. Interestingly, we noted relatively high odds of ileostomy or colostomy among non-MMC patients. While the exact reason for this finding is unclear, we suspect that our results are influenced by the presence of non-MMC patients with other co-morbid diagnoses such as cloacal exstrophy or anorectal malformations.

As these studies highlight, the decisions to proceed with surgical management of neurogenic bowel issues in SB patients are complex and involve many aspects of patients' medical condition. We noted a similar variation in surgical management rates of neurogenic bladder among NSBPR clinics [19], and the pattern is also apparent in the choice of procedure since ACE/cecostomy procedures are commonly performed with bladder reconstruction. Importantly, those decisions also seem to include consideration of other non-medical aspects of each SB patient.

Given the complexity of surgical decision-making in this population, it is perhaps unsurprising that some variation should exist among clinics caring for individuals with SB. Variation is likely a reflection, in part, of surgeon philosophy. Surgical variation may be further influenced by the population, social situation and the level of support and resources available at home and in the local environment. These factors are not necessarily reflected by the demographic or socioeconomic data available in the NSBPR. Another contributing factor could be allocation of limited resources (e.g., surgeon time) and possible lower reimbursement potential for longer reconstructive cases, particularly given high rates of public insurance coverage among these patients.

Regardless of the root causes, this degree of variation is intriguing as it implies either overuse or under-use of these procedures; such variation can be problematic [20–23]. A high degree of variation in the use of these procedures is of interest to clinicians, researchers, and policy makers alike, as such variation implies that there is not an obvious or widely accepted standard of care for the management of neurogenic bowel, including surgical management. Further, such variation typically implies either overuse or un-deruse of these procedures, either of which may be a significant problem in medically complex patients. Overuse of neurogenic bowel procedures is of concern due to the significant potential morbidity and expense of surgery; underuse is problematic because constipation and fecal incontinence can

have a detrimental impact on patients' quality of life and may predispose to other health concerns.

Our results suggest that the hospital at which the individual is treated plays a role in surgical decision-making along with clinical factors such as age, race, mobility status, and lesion level. Given the significant social and financial costs of neurogenic bowel procedures, this level of variation suggests that further research is needed to help clinicians and patients make appropriate and less arbitrary choices regarding surgery.

The findings of our study must be interpreted in the context of study limitations. The NSBPR continues to undergo improvements and modifications to ensure the validity of its data. Whereas clear definitions are provided for the functional outcomes that we have analyzed, these are still potentially subject to variation in their interpretation and reporting by different individuals at different clinics. This raises particular caveats when attempting to compare outcomes from different clinics. As with any large database, it is also possible that there are errors in data acquisition and input. However, previous NSBPR studies have shown that these errors should be infrequent [8].

Selection bias may be a threat to external validity. NSBPR is clinic-based, so it may not represent SB patients who do not attend SB clinics. It is also possible that the clinics participating in the registry are not representative of SB clinics in general. No attempt was made to ensure representativeness in choosing NSBPR clinics; indeed, the fact that all NSBPR clinics are multidisciplinary may limit generalizability of our findings. Nevertheless, we believe the NSBPR probably characterizes the majority of SB clinics in the United States. In addition, selection bias may be a threat to internal validity: participating clinics enrolled most but not all eligible patients, raising concerns that those who are eligible but not contributing data may be different from those who are. In an attempt to evaluate the possible impact of this bias, we conducted a rigorous selection bias analysis (Table 5) based on the best available literature on this topic. Importantly, we did not see a significant difference in outcomes before and after these adjustments, implying that selection bias may not play a significant role in our findings.

5. Conclusions

There is significant variation in neurogenic bowel procedure rates among NSBPR clinics. In addition to SB-related factors such as bladder surgery, mobility status and lesion type/level, non-SB-related factors such as patient age, gender, race/ethnicity and treating center are also associated with the likelihood of undergoing neurogenic bowel procedures.

Acknowledgments

The development of the National Spina Bifida Patient Registry has been successful due to the contributions of all the members of the NSBPR Coordinating Committee. Members of this committee during the collection of the data reported here were William Walker, Seattle Children's Hospital; Kathryn Smith, Children's Hospital, Los Angeles; Kurt Freeman, Oregon Health and Science University; Pamela Wilson, Children's Hospital Colorado; Kathleen Sawin, Children's Hospital of Wisconsin and Froedtert Hospital, Milwaukee (adult clinic); Jeffrey Thomson, Connecticut Children's Medical Center, and Shriners Hospital for Children, Springfield; Heidi Castillo, Children's Hospital Medical Center, Cincinnati, and Texas Children's Hospital, Houston; David Joseph, Children's Hospital of Alabama; Jacob Neufeld, St. Luke's Boise Medical Center, Boise; Robin Bowman, Lurie Children's Hospital of

Chicago, Chicago; Karen Ratliff-Schaub, Nationwide Children's Hospital, Columbus; Jim Chinarian, Children's Hospital of Michigan, Detroit; John Wiener, Duke University Medical Center, Durham; Mark Dias, Hershey Medical Center, Hershey; Joe O'Neil, Riley Hospital for Children, Indianapolis; Alex VanSpeybroeck, Shriners Hospital for Children, Los Angeles; Brad Dicianno, Children's Hospital of Pittsburgh, and University of Pittsburgh Medical Center (adult clinic), Pittsburgh; Paula Peterson, Primary Children's Medical Center, Salt Lake City; Elaine Pico, UCSF, San Francisco, and Children's Hospital and Research Center, Oakland; Nienke Dosa, Upstate Golisano Children's Hospital, Syracuse; Stacy Tanaka, Monroe Jr. Carrell Children's Hospital at Vanderbilt, Nashville; Carlos Estrada, Boston Children's Hospital, Boston; Michael Partington, Gillette Children's Specialty Healthcare, St. Paul.

This project is supported by the Centers for Disease Control and Prevention (CDC) for the National Spina Bifida Patient Registry (U01-DD001087). Dr. Routh is also supported in part by grant K08-DK100534 from the National Institute of Diabetes and Digestive and Kidney Diseases (NIDDK). The funding sources had no role in the collection, analysis or interpretation of data; in the writing of the manuscript; or in the decision to submit the manuscript for publication. The findings and conclusions in this report are those of the authors and do not necessarily represent the official position of the Centers for Disease Control and Prevention.

References

1. Lloyd JC, Wiener JS, Gargollo PC, et al. Contemporary epidemiological trends in complex congenital genitourinary anomalies. J Urol. 2013; 190:1590. [PubMed: 23791903]
2. Parker SE, Mai CT, Canfield MA, et al. Updated National Birth Prevalence estimates for selected birth defects in the United States, 2004–2006. Birth Defects Res A Clin Mol Teratol. 2010; 88:1008. [PubMed: 20878909]
3. Snow-Lisy DC, Yerkes EB, Cheng EY. Update on Urological Management of Spina Bifida from Prenatal Diagnosis to Adulthood. J Urol. 2015
4. Szymanski, K., Misseri, R., Whittam, B., et al. Impact of urinary and fecal incontinence on quality of life and in adults with spina bifida: all incontinence is not created equal. Presented at the Society for Pediatric Urology Annual Meeting; San Diego, CA. 2016;
5. Scales CD Jr, Wiener JS. Evaluating outcomes of enterocystoplasty in patients with spina bifida: a review of the literature. J Urol. 2008; 180:2323. [PubMed: 18930285]
6. Wiener JS, Antonelli J, Shea AM, et al. Bladder augmentation versus urinary diversion in patients with spina bifida in the United States. J Urol. 2011; 186:161. [PubMed: 21575969]
7. Sawin KJ, Liu T, Ward E, et al. The National Spina Bifida Patient Registry: profile of a large cohort of participants from the first 10 clinics. J Pediatr. 2015; 166:444. [PubMed: 25444012]
8. Thibadeau JK, Ward EA, Soe MM, et al. Testing the feasibility of a National Spina Bifida Patient Registry. Birth Defects Res A Clin Mol Teratol. 2013; 97:36. [PubMed: 23125114]
9. Schechter MS, Liu T, Soe M, et al. Sociodemographic attributes and spina bifida outcomes. Pediatrics. 2015; 135:e957. [PubMed: 25780069]
10. Maclure M, Hankinson S. Analysis of selection bias in a case-control study of renal adenocarcinoma. Epidemiology. 1990; 1:441. [PubMed: 2090281]
11. Lash, TL., Fox, MP., Fink, AK. Applying Quantitative Bias Analysis to Epidemiologic Data. New York, NY: Springer; 2009. Selection Bias; p. 117-150.
12. Malone PS, Ransley PG, Kiely EM. Preliminary report: the antegrade continence enema. Lancet. 1990; 336:1217. [PubMed: 1978072]
13. Christison-Lagay ER, Rodriguez L, Kurtz M, et al. Antegrade colonic enemas and intestinal diversion are highly effective in the management of children with intractable constipation. J Pediatr Surg. 2010; 45:213. [PubMed: 20105606]
14. Chu DI, Balsara ZR, Routh JC, et al. Experience with glycerin for antegrade continence enema in patients with neurogenic bowel. Journal of Urology. 2013; 189:690. [PubMed: 22986031]
15. Myers JB, Hu EM, Elliott SP, et al. Short-term outcomes of Chait Trapdoor for antegrade continence enema in adults. Urology. 2014; 83:1423. [PubMed: 24703460]
16. Corbett P, Denny A, Dick K, et al. Peristeen integrated transanal irrigation system successfully treats faecal incontinence in children. J Pediatr Urol. 2014; 10:219. [PubMed: 24439630]
17. Imai K, Shiroyanagi Y, Kim WJ, et al. Satisfaction after Malone antegrade continence enema procedure in patients with spina bifida. Spinal Cord. 2014; 52:54. [PubMed: 24081017]

18. Teichman JM, Harris JM, Currie DM, et al. Malone antegrade continence enema for adults with neurogenic bowel disease. Journal of Urology. 1998; 160:1278. [PubMed: 9751335]
19. Routh JC, Joseph DB, Liu T, et al. Bladder Reconstruction Rates Differ Among Centers Participating in National Spina Bifida Patient Registry. Journal of Urology. 2017 In press.
20. Kleinman LC, Kosecoff J, Dubois RW, et al. The medical appropriateness of tympanostomy tubes proposed for children younger than 16 years in the United States. JAMA. 1994; 271:1250. [PubMed: 7710469]
21. Lieu TA, Lozano P, Finkelstein JA, et al. Racial/ethnic variation in asthma status and management practices among children inmanaged medicaid. Pediatrics. 2002; 109:857. [PubMed: 11986447]
22. Perrin JM, Homer CJ, Berwick DM, et al. Variations in rates of hospitalization of children in three urban communities. N Engl J Med. 1989; 320:1183. [PubMed: 2710191]
23. Routh JC, Nelson CP, Graham DA, et al. Variation in surgical management of vesicoureteral reflux: influence of hospital and patient factors. Pediatrics. 2010; 125:e446. [PubMed: 20142292]

Table 1

Characteristics of included patients, NSBPR, 2009–2014

	N (%) or statistics by bowel procedure			
Variables	**Overall n (%) (N = 5528)**	**Yes (*n* = 1088)**	**No (*n* = 4440)**	**P-value**
Age group at last visit (yrs)				< 0.001
Younger than 2	474 (8.6)	10 (2.1)	464 (97.9)	
2 to < 5	716 (13.0)	25 (3.5)	691 (96.5)	
5 to < 10	1219 (22.1)	184 (15.1)	1035 (84.9)	
10 to < 13	627 (11.3)	161 (25.7)	466 (74.3)	
13 to < 18	1034 (18.7)	317 (30.7)	717 (69.3)	
18 to < 22	656 (11.9)	209 (31.9)	447 (68.1)	
22 or older	802 (14.5)	182 (22.7)	620 (77.3)	
Sex				0.006
Male	2616 (47.3)	474 (18.1)	2142 (81.9)	
Female	2912 (52.7)	614 (21.1)	2298 (78.9)	
Race/ethnicity				< 0.001
Non-Hispanic white	3542 (64.1)	823 (23.2)	2719 (76.8)	
Non-Hispanic black	400 (7.2)	65 (16.3)	335 (83.8)	
Hispanic or Latino	1167 (21.1)	142 (12.2)	1025 (87.8)	
Other/refused/unknown	419 (7.6)	58 (13.8)	361 (86.2)	
Spina bifida type				< 0.001
Myelomeningocele	4393 (79.5)	958 (21.8)	3435 (78.2)	
Non-myelomeningocele	1135 (20.5)	130 (11.5)	1005 (88.5)	
Function level of lesion				< 0.001
Thoracic	864 (15.6)	268 (31.0)	596 (69.0)	
Lumbar	2957 (53.5)	621 (21.0)	2336 (79.0)	
Sacral	1707 (30.9)	199 (11.7)	1508 (88.3)	
Health insurance				< 0.001
Any private	2604 (47.1)	579 (22.2)	2025 (77.8)	
Non-private	2923 (52.9)	509 (17.4)	2414 (82.6)	
Mobility status				< 0.001
Community ambulators	2960 (53.6)	463 (15.6)	2497 (84.4)	
Household ambulators	395 (7.1)	92 (23.3)	303 (76.7)	
Therapeutic ambulators	393 (7.1)	93 (23.7)	300 (76.3)	
Non-ambulators	1696 (30.7)	437 (25.8)	1259 (74.2)	
Clinic				< 0.001
1	550 (9.9)	94 (17.1)	456 (82.9)	
2	93 (1.7)	9 (9.7)	84 (90.3)	
3	87 (1.6)	8 (9.2)	79 (90.8)	
4	224 (4.1)	62 (27.7)	162 (72.3)	
5	350 (6.3)	70 (20.0)	280 (80.0)	
6	314 (5.7)	67 (21.3)	247 (78.7)	

	N (%) or statistics by bowel procedure			
Variables	**Overall n (%) (N = 5528)**	**Yes (*n* = 1088)**	**No (*n* = 4440)**	**P-value**
7	341 (6.2)	97 (28.4)	244 (71.6)	
8	330 (6.0)	126 (38.2)	204 (61.8)	
9	280 (5.1)	40 (14.3)	240 (85.7)	
10	384 (6.9)	27 (7.0)	357 (93.0)	
11	315 (5.7)	34 (10.8)	281 (89.2)	
12	457 (8.3)	73 (16.0)	384 (84.0)	
13	414 (7.5)	99 (23.9)	315 (76.1)	
14	396 (7.2)	67 (16.9)	329 (83.1)	
15	182 (3.3)	27 (14.8)	155 (85.2)	
16	344 (6.2)	108 (31.4)	236 (68.6)	
17	92 (1.7)	21 (22.8)	71 (77.2)	
18	298 (5.4)	57 (19.1)	241 (80.9)	
19	77 (1.4)	2 (2.6)	75 (97.4)	

Table 2

Bivariate analysis of the probability of undergoing any neurogenic bowel procedure, NSBPR (2009–2014)

Variables	N (%) (Total N = 5528)	Odds ratio (95% CI)	P-value
Age group at last visit (years)			< 0.001
Younger than 2 †	474 (8.6)	Reference	
2 to < 5	716 (13.0)	1.68 (0.80–3.53)	
5 to < 10	1219 (22.1)	8.25 (4.32–15.73)	
10 to < 13	627 (11.3)	16.03 (8.36–30.75)	
13 to < 18	1034 (18.7)	20.51 (10.81–38.91)	
18 to < 22	656 (11.9)	21.69 (11.35–41.46)	
22 or older	802 (14.5)	13.62 (7.13–26.03)	
Gender			0.006
Male †	2616 (47.3)	Reference	
Female	2912 (52.7)	1.21 (1.06–1.38)	
Race			< 0.001
Non-Hispanic white †	3542 (64.1)	Reference	
Non-Hispanic black	400 (7.2)	0.64 (0.49–0.85)	
Hispanic or Latino	1167 (21.1)	0.46 (0.38–0.55)	
Other/refused/unknown	419 (7.6)	0.53 (0.40–0.71)	
Spina bifida type			< 0.001
Myelomeningocele †	4393 (79.5)	Reference	
Non-myelomeningocele	1135 (20.5)	0.46 (0.38–0.56)	
Functional level of lesion			< 0.001
Thoracic †	864 (15.6)	Reference	
Lumbar	2957 (53.5)	0.59 (0.50–0.70)	
Sacral	1707 (30.9)	0.29 (0.24–0.36)	
Insurance			< 0.001
Any private †	2604 (47.1)	Reference	
Non-private	2923 (52.9)	0.74 (0.65–0.84)	
Mobility status			< 0.001
Community ambulators †	2960 (53.6)	Reference	
Household ambulators	395 (7.1)	1.64 (1.27–2.11)	
Therapeutic ambulators	393 (7.1)	1.67 (1.30–2.15)	
Non-ambulators	1696 (30.7)	1.87 (1.62–2.17)	
Clinic			< 0.001
1	550 (9.9)	Reference	
2	93 (1.7)	0.52 (0.25–1.07)	
3	87 (1.6)	0.49 (0.23–1.05)	
4	224 (4.1)	1.86 (1.29–2.68)	
5	350 (6.3)	1.21 (0.86–1.71)	
6	314 (5.7)	1.32 (0.93–1.87)	

Variables	N (%) (Total N = 5528)	Odds ratio (95% CI)	P-value
7	341 (6.2)	1.93 (1.40–2.67)	
8	330 (6.0)	3.00 (2.19–4.10)	
9	280 (5.1)	0.81 (0.54–1.21)	
10	384 (6.9)	0.37 (0.23–0.58)	
11	315 (5.7)	0.59 (0.39–0.89)	
12	457 (8.3)	0.92 (0.66–1.29)	
13	414 (7.5)	1.52 (1.11–2.09)	
14	396 (7.2)	0.99 (0.70–1.39)	
15	182 (3.3)	0.85 (0.53–1.35)	
16	344 (6.2)	2.22 (1.62–3.05)	
17	92 (1.7)	1.43 (0.84–2.45)	
18	298 (5.4)	1.15 (0.80–1.65)	
19	77 (1.4)	0.13 (0.03–0.54)	

[†]Reference group.

Table 3

Multivariable analysis of the probability of undergoing any neurogenic bowel procedure, NSBPR (2009–2014)

Variables	Adjusted odds ratio (95% CI)	P-value
Age group at annual visit		< 0.001
Younger than 2	Reference	
2 to < 5	2.45 (1.04–5.78)	
5 to < 10	12.73 (5.84–27.76)	
10 to < 13	27.87 (12.72–61.11)	
13 to < 18	31.13 (14.37–67.43)	
18 to < 22	34.06 (15.60–74.36)	
22+	16.46 (7.51–36.09)	
Sex		0.015
Male	Reference	
Female	1.20 (1.04–1.39)	
Race/ethnicity		0.002
Non-Hispanic white	Reference	
Non-Hispanic black	0.79 (0.58–1.07)	
Hispanic or Latino	0.64 (0.50–0.81)	
Other	0.79 (0.57–1.08)	
Spina bifida type		0.012
Myelomeningocele	Reference	
Non-myelomeningocele	0.74 (0.58–0.94)	
Function level of lesion		< 0.001
Thoracic	Reference	
Lumbar	0.82 (0.65–1.03)	
Sacral	0.54 (0.40–0.74)	
Mobility status		0.011
Community ambulators	Reference	
Household ambulators	1.40 (1.04–1.88)	
Therapeutic ambulators	1.35 (1.00–1.81)	
Non-ambulators	1.43 (1.14–1.80)	
Insurance		0.002
Any private	Reference	
Non-private	0.78 (0.67–0.91)	
Clinic		< 0.001
1	Reference	
2	0.54 (0.26–1.14)	
3	0.46 (0.21–1.01)	
4	2.02 (1.36–3.02)	
5	1.65 (1.14–2.40)	
6	1.57 (1.07–2.30)	
7	2.61 (1.82–3.74)	

Variables	Adjusted odds ratio (95% CI)	P-value
8	4.23 (2.97–6.04)	
9	1.44 (0.93–2.23)	
10	0.54 (0.33–0.90)	
11	0.57 (0.37–0.89)	
12	1.31 (0.91–1.88)	
13	1.63 (1.16–2.29)	
14	1.17 (0.80–1.70)	
15	0.86 (0.53–1.41)	
16	2.38 (1.68–3.36)	
17	2.51 (1.38–4.58)	
18	1.53 (1.04–2.26)	
19	0.20 (0.05–0.84)	

Table 4

Bivariate and multiple logistic regression models of the probability of undergoing either ileostomy or colostomy among patients undergoing neurogenic bowel procedure, NSBPR, 2009–2014

Variables	N (%) (Total N = 1064)	Crude odds ratio (95% CI)	Adjusted odds ratio (95% CI)	P-value
Age group at annual visit				< 0.001
2 to < 5	25 (2.4)	Reference	Reference	
5 to < 10	181 (17.2)	0.08 (0.03–0.21)	0.10 (0.03–0.37)	
10 to < 13	154 (14.6)	0.05 (0.02–0.14)	0.05 (0.01–0.21)	
13 to < 18	309 (29.3)	0.02 (0.01–0.06)	0.02 (0.00–0.07)	
18 to < 22	206 (19.5)	0.04 (0.01–0.10)	0.02 (0.01–0.09)	
22+	179 (17.0)	0.11 (0.04–0.27)	0.10 (0.03–0.39)	
Race/ethnicity				0.004
Non-Hispanic white	805 (75.7)	Reference	Reference	
Non-Hispanic black	64 (6.0)	3.06 (1.66–5.65)	4.57 (1.91–10.95)	
Hispanic or Latino	139 (13.1)	2.32 (1.44–3.73)	0.93 (0.38–2.23)	
Other	56 (5.3)	1.53 (0.70–3.35)	0.61 (0.21–1.79)	
Spina bifida type				< 0.001
Myelomeningocele	941 (88.4)	Reference	Reference	
Non-myelomeningocele	123 (11.6)	10.12 (6.61–15.50)	23.97 (11.11–51.73)	
Mobility status				< 0.001
Community ambulators	450 (42.4)	Reference	Reference	
Household ambulators	91 (8.6)	0.74 (0.34–1.61)	1.83 (0.56–5.96)	
Therapeutic ambulators	91 (8.6)	0.64 (0.28–1.45)	1.41 (0.45–4.37)	
Non-ambulators	429 (40.4)	1.27 (0.85–1.89)	7.07 (3.40–14.70)	
Any genitourinary surgery				< 0.001
Yes	622 (58.5)	Reference	Reference	
No	442 (41.5)	4.86 (3.22–7.34)	4.53 (2.65–7.76)	
Clinic *				< 0.001
1	91 (8.6)	Reference	Reference	
2	9 (0.8)	0.51 (0.06–4.32)	*	
3	8 (0.8)	1.35 (0.25—7.26)	2.48 (0.33–18.50)	
4	62 (5.8)	0.36 (0.12–1.02)	0.22 (0.05–1.03)	
5	66 (6.2)	1.09 (0.50–2.39)	0.82 (0.30–2.26)	
6	65 (6.1)	0.20 (0.06–0.70)	0.31 (0.07–1.30)	
7	97 (9.1)	0.09 (0.02–0.38)	0.03 (0.00–0.40)	
8	126 (11.9)	0.17 (0.06–0.47)	0.24 (0.06–0.90)	
9	40 (3.8)	0.21 (0.05–0.97)	0.22 (0.04–1.28)	
10	27 (2.5)	3.24 (1.30–8.12)	8.37 (1.94–36.12)	
11	33 (3.1)	1.30 (0.50–3.35)	1.31 (0.39–4.42)	
12	70 (6.6)	1.62 (0.78–3.37)	1.50 (0.50–4.55)	
13	99 (9.3)	0.26 (0.10–0.69)	0.15 (0.05–0.50)	
14	66 (6.2)	0.56 (0.23–1.38)	0.58 (0.16–2.08)	

Variables	N (%) (Total N = 1064)	Crude odds ratio (95% CI)	Adjusted odds ratio (95% CI)	P-value
15	25 (2.4)	1.01 (0.33–3.07)	1.11 (0.27–4.57)	
16	105 (9.9)	0.38 (0.16–0.89)	0.30 (0.10–0.90)	
17	21 (2.0)	0.43 (0.09–2.00)	0.38 (0.04–3.45)	
18	52 (4.9)	0.63 (0.24–1.63)	0.47 (0.14–1.56)	

* Clinics with extreme surgery distributions were excluded from this analysis.

Table 5

Summary of selection bias analysis for 3,067 neurogenic bowel procedures, NSBPR, 2014

Variables	SPR, median (2.5, 97.5 percentiles)^		Estimate of bias (median RSPR*)	Observed odds ratio (95% CI)**	Adjusted odds ratio (95% CI)
	Surgery	No surgery			
	Odds of enrollment compared with referent group		Likelihood of being enrolled if the patient had surgery	Odds of having surgery compared with the reference group	Odds of having surgery compared with the reference group adjusted for enrollment
Age (years)					
< 10†	Referent	Referent		Referent	Referent
10 to < 18	1.52 (0.86, 2.54)	1.48 (1.32, 1.69)	1.02	4.28 (3.37, 5.44)	4.19 (3.30, 5.32)
18 or older	0.73 (0.41, 1.20)	0.85 (0.76, 0.97)	0.86	3.10 (2.35, 4.08)	3.61 (2.74, 4.76)
Sex					
Male†	Referent	Referent		Referent	Referent
Female	1.20 (0.82, 1.76)	1.07 (0.99, 1.16)	1.12	1.18 (0.96, 1.44)	1.05 (0.86, 1.28)
Race/ethnicity					
NH white†	Referent	Referent		Referent	Referent
NH black	0.77 (0.42, 1.87)	0.68 (0.61, 0.78)	1.13	0.74 (0.48, 1.11)	0.65 (0.43, 0.99)
Hispanic or Latino	1.01 (0.60, 2.01)	1.09 (1.00, 1.20)	0.93	0.53 (0.40, 0.72)	0.57 (0.43, 0.77)
Other	1.03 (0.57, 2.55)	1.70 (1.44, 2.11)	0.61	1.05 (0.72, 1.52)	1.72 (1.18, 2.49)
Diagnosis					
Myelomeningocele†	Referent	Referent		Referent	Referent
Other diagnosis	0.62 (0.36, 1.22)	0.54 (0.49, 0.59)	1.15	0.70 (0.51, 0.96)	0.61 (0.44, 0.84)
Level of lesion					
Thoracic†	Referent	Referent		Referent	Referent
Lumbar	1.16 (0.75, 1.75)	1.20 (1.01, 1.38)	0.97	0.72 (0.55, 0.93)	0.74 (0.57, 0.96)
Sacral	1.25 (0.70, 2.53)	1.18 (0.99, 1.38)	1.06	0.41 (0.29, 0.58)	0.39 (0.27, 0.54)
Insurance					
Private†	Referent	Referent		Referent	Referent
Non-private	0.40 (0.26, 0.59)	0.46 (0.42, 0.51)	0.86	0.77 (0.62, 0.95)	0.89 (0.72, 1.10)

*Ratio of selection probability ratio.

^ Simulation of assigning surgery status for eligible but not enrolled patients was executed 10,000 times.

** Eligible and enrolled patients.

† Reference group.