Neurogenic bowel treatments and continence outcomes in children and adults with myelomeningocele

Maryellen S. Kelly[a,*], **John S. Wiener**[a,1], **Tiebin Liu**[b], **Priya Patel**[c], **Heidi Castillo**[d], **Jonathan Castillo**[d], **Brad E. Dicianno**[e], **Joan Jasien**[f], **Paula Peterson**[g], **Jonathan C. Routh**[a], **Kathleen Sawin**[h], **Eileen Sherburne**[h], **Kathryn Smith**[i], **Asma Taha**[j], **Gordon Worley**[f,1,2]

[a]Division of Urology, Department of Surgery, Duke University, Durham, NC, USA

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

[c]Oak Ridge Institute for Science and Education (ORISE), Oak Ridge, TN, USA

[d]Department of Pediatrics, Baylor College of Medicine, Houston, TX, USA

[e]Department of Physical Medicine and Rehabilitation, University of Pittsburgh School of Medicine, Pittsburgh, PA, USA

[f]Division of Pediatric Neurology, Department of Pediatrics, Duke University, Durham, NC, USA

[g]Department of Pediatrics, University of Utah, Salt Lake City, UT, USA

[h]Department of Nursing Research, Children's Hospital of Wisconsin, USA and Self-management Science Center, College of Nursing, University of Wisconsin-Milwaukee, Milwaukee, WI, USA

[i]Department of Pediatrics, Keck School of Medicine, Los Angeles, CA, USA

[j]Doernbecher Children's Hospital and School of Nursing, Oregon Health Sciences University, Portland, OR, USA

Abstract

PURPOSE: Neurogenic bowel dysfunction (NBD) is a common comorbidity of myelomeningocele (MMC), the most common and severe form of spina bifida. The National

Spina Bifida Patient Registry (NSBPR) is a research collaboration between the CDC and Spina Bifida Clinics. Fecal continence (continence) outcomes for common treatment modalities for NBD have not been described in a large sample of individuals with MMC. NSBPR patients with MMC and NBD were studied to determine variation in continence status and their ability to perform their treatment independently according to treatment modality and individual characteristics.

METHODS: Continence was defined as < 1 episode of incontinence per month. Eleven common treatments were evaluated. Inclusion criteria were established diagnoses of both MMC and NBD, as well as age   5 years (n = 3670). Chi-square or exact statistical tests were used for bivariate analyses. Logistic regression models were used to estimate the odds of continence outcomes by age, sex, race/ethnicity, level of motor function, and insurance status.

RESULTS: At total of 3670 members of the NSBPR met inclusion criteria between November 2013 and December 2017. Overall prevalence of continence was 45%. Prevalence ranged from 40–69% across different treatments. Among continent individuals, 60% achieved continence without surgery. Antegrade enemas were the most commonly used treatment and had the highest associated continence rate. Ability to carry out a treatment independently increased with age. Multivariable logistic regression showed significantly higher odds of continence among individuals aged   12 years, female, non-Hispanic white, and with private insurance.

CONCLUSIONS: The prevalence of continence was generally low and varied between treatment modalities. Better treatment algorithms considering patient factors may result in improved outcomes.

Keywords

1. Introduction

Myelomeningocele (MMC) is the most common and severe form of spina bifida (SB) and is a permanently disabling congenital condition. Neurogenic bowel dysfunction (NBD) is a significant comorbidity that affects most individuals with MMC, resulting in fecal incontinence and/or chronic constipation. Fecal incontinence in MMC has been associated with decreased health-related quality of life, depression, discrimination by peers, decreased school attendance, lower educational attainment, and lower rates of employment [1–3]. Worrying about fecal continence contributes to emotional, physical, and psychological distress [4]. Families and caregivers of individuals with NBD report decreased quality of life, the causes of which include the time, effort, and unpleasantness of helping with NBD management [5–9]. Secondary concerns from NBD include urinary incontinence, urinary tract infections, ventriculoperitoneal shunt malfunction, skin breakdown, hemorrhoids, and anal fissures [10–16]. Of MMC individuals with NBD, 80% are on a management program for constipation and/or fecal continence [17,18].

The National Spina Bifida Patient Registry (NSBPR) is a collaboration between the CDC and 35 clinics across the U.S. The purpose of the NSBPR is to provide a research platform to improve the care of people with SB. Data about fecal continence status and treatments used for NBD are collected annually. In the initial report from the NSBPR from 10 clinics,

only 30% of individuals with MMC reported fecal continence. This was reported as a binary subjective report of continence [19]. Later, the prevalence of continence ranged from 24% to 59% among 12 clinics with more than 200 individuals enrolled. This variation suggests that there may be modifiable factors that could improve NBD outcomes. Additionally, prior studies with the NSBPR have shown that sociodemographic factors may be related to fecal continence rates [20].

To date, there is no standard of care for the management of NBD and its associated outcomes including fecal incontinence and constipation. Therefore, we used data from the NSBPR to compare continence outcomes across common treatment modalities for NBD and to identify sociodemographic factors associated with these outcomes and the individuals' ability to perform their own treatment independently.

2. Methods

2.1. NSBPR

Standard data collection tools are used to obtain de-identified data from each of the multidisciplinary SB clinics. After initial enrollment, an annual visit form is used to update information on demographic and clinical characteristics as well as treatment history. Clinics can collect data for the visit form in various ways, including face-to-face interviews, written survey instruments, or clinician notes. Data collection for the registry was approved by the Institutional Review Board of each clinic site. To avoid diagnostic heterogeneity, our sample was limited to those with MMC.

Data from the most recent clinic visit were analyzed. Inclusion criteria were: MMC form of SB, last visit after October 2013 (when data collection forms were revised to include frequency of fecal incontinence episodes), age 5 years at last visit, and presence of NBD. We defined fecal continence as having episodes of incontinence less than once a month or never. Although NBD is often a combination of symptoms of constipation and/or fecal incontinence, the NSBPR does not capture data on constipation. Therefore this study only assessed the symptoms of fecal incontinence secondary to NBD. We defined fecal incontinence secondary to NBD as having one or more episodes of fecal incontinence per month and/or using any treatment modality for NBD. Those selecting "cannot assess" in regards to fecal continence were excluded ($n = 229$). Those who reported managing their condition with pouched fecal diversion, ileostomy, or colostomy were excluded from continence analyses.

2.2. Sociodemographic characteristics

Age was categorized by: early childhood (5–11 years), adolescent (12–19 years), and adult (over 19 years of age). Health insurance status was recorded at each visit as either having any private insurance or no private insurance. Sex and race/ethnicity data were also collected.

2.3. Spinal cord level of motor function

The lowest spinal cord level of motor function found on examination was recorded for each lower limb as: thoracic (flaccid lower limb); high-lumbar (hip flexion present); mid-lumbar (knee extension present); low-lumbar (foot dorsiflexion present); or sacral (foot plantar flexion present). When the left and right sides differed, the more severely involved side was used to represent that individual's overall functional level.

2.4. NBD management techniques and treatment modalities

Data were collected on 11 frequently used treatment modalities for NBD (Table 1); the online Supplementary Appendix B has more detailed information about these modalities). As a construct for the analyses, criterion treatment modalities (CTMs) for NBD were ranked based on invasiveness of administration. Treatment options requiring surgery were judged to be more invasive than non-surgical options. When a person reported more than one CTM, the most invasive treatment modality was used to classify that person's CTM. The continence rates associated with using a CTM exclusively versus in conjunction with other CTMs was analyzed.

2.5. Ability to perform CTM independently

CTM data for each individual was collected as a binary variable regarding their ability to perform their own CTM independently.

2.6. Statistical analysis

Associations among independent categorical variables were evaluated by chi-square tests. When an expected cell count was   5, the Fisher's exact method was used to test associations. Univariate and multiple logistic regression models were used to test associations of continence outcomes with sex of participants, level of motor function, health insurance status, and CTM. Statistical tests were all 2-sided, and p values < 0.05 were considered significant.

Individuals were stratified by motor function category. Univariate logistic regression models were used to determine the odds of continence for each CTM, compared to timed defecation (referent). Timed defecation was used as referent because it is the least invasive CTM. Statistical analyses were performed using SAS version 9.4 (Cary, NC, USA).

3.   Results

3.1.   Study population

Through December 2017, 8662 individuals were enrolled in the NSBPR and 3670 met our inclusion criteria and composed our study population (Fig. 1). Demographic and clinical characteristics are shown in Table 2. Similar to other NSBPR studies, this study had high proportions of females (56.2% among those 20 years old or older), non-Hispanic whites (63% overall), individuals with mid-lumbar level lesions (31.6% overall), and individuals with no private insurance (57.1%) [3,20,21].

3.2. Use of CTMs

Rates of usage of each CTM is presented in Table 3. The most commonly used CTM in this population with NBD was antegrade enemas (27%), followed by oral medications only (23%). The prevalence of use of each CTM by age category and by level of motor function is presented in online Appendix Tables A1 and A2. The factors that increased the odds of using any CTM were adolescent age, female sex, non-Hispanic ethnicity, and having private insurance (see online Appendix Table A3).

3.3. Fecal continence outcomes

The overall prevalence of fecal continence in the study population was 45%. Among those that were continent, 60% had no record of surgical intervention. Additionally, 14.1% of the study population did not report any CTM and were incontinent.

On multiple logistic regression analysis, higher odds of continence were associated with being older, female, non-Hispanic white, or having any private health insurance. Individuals with sacral and low-lumbar level of motor function also had higher odds of continence than those with higher levels of motor function involvement (Table 4).

3.4. Fecal continence outcomes by CTM

The overall rate of continence associated with each CTM ranged from 35.9% (manual disimpaction) to 68.6% (antegrade enemas) (Table 3). Antegrade enemas were also the most commonly used CTM (26.5%). Cone/balloon large volume enemas and Peristeen® transanal irrigation were associated with the second highest overall continence rates (58.5% and 57.9%, respectively).

The prevalence of continence increased with age for all CTMs except cone/balloon enemas and mini-enema, for which peak continence rates were found in adolescents. In children, antegrade enemas had the highest associated continence rate (66%), followed by cone/balloon enemas (57%). In adolescents, transanal irrigation had the highest associated continence rate (80%), while antegrade enemas had the second highest rate (70%). In adults, timed defecation had the highest associated continence rate (79%), followed by standard rectal enemas (76%) (Table 3).

The variation in continence rates by level of motor function was statistically significant for only two of the CTMs: oral medications and rectal suppositories. These two CTMs both showed higher rates of continence in individuals with low lumbar and sacral levels than those with higher levels (Appendix Table A4).

Univariate logistic regression analysis for continence with a CTM by level of motor function found that antegrade enemas were significantly associated with continence in each level of motor function (Table A5). Retrograde large volume enemas (combined Cone/balloon large volume enemas and Peristeen® transanal irrigation) were significantly associated with continence only in those with high-lumbar lesions (odds ratio 3.0, 95% confidence interval 1.44–6.26, p-value = 0.0034) (Appendix Table A5).

3.5. Ability to independently perform CTM

Only 23% of individuals were independently able to perform their own CTM. The overall rate of independence in carrying out a CTM ranged from 13% for transanal irrigation to 56% for antegrade enemas. The rate of independent use for each CTM increased with age (Table 5). Timed defecation was the CTM with the highest probability of being carried out independently in all age groups. Most individuals, regardless of age, were not able to administer other CTMs independently.

3.6. Combinations of CTMs

The continence rates for each CTM when used on its own or in conjunction with an oral agent are listed in Table 6. The addition of oral agents to a CTM was associated with a lower rate of continence for all CTMs, which was statistically significant for six of the nine CTMs examined.

Only 5.5% of individuals utilized more than one CTM (177 used 2 CTMs, 21 used 3 CTMs, 3 used 4 CTMs, and 2 used 5 CTMs). Over half (53%) of those who used 2 or more CTMs had timed defecation as their second CTM. When a CTM was used in conjunction with another CTM, the continence rate did not increase, except for manual disimpaction (see online Appendix Table A6).

4. Discussion

To our knowledge, this is the first study to systematically evaluate the use and effectiveness of common treatment modalities for NBD in individuals with MMC. The goal of NBD treatment is to find the most effective and least intrusive management for each individual. The use of a large multi-institutional cohort allowed acquisition of sufficient statistical power to allow meaningful comparisons between CTMs.

The fecal continence rate for the overall actively managed cohort was low at 45%. Of this MMC cohort with NBD, 14% did not report using any CTM and reported being incontinent. Given the association of fecal continence with employment and educational attainment for adults with MMC [3], we believe this overall low fecal continence rate to be a major problem for this population.

Our definition of fecal continence was having less than one fecal incontinent episode per month. Some individuals may have more than one incontinent episode but are satisfied with their continence; satisfaction was not directly evaluated in this study. Furthermore, the registry does not collect the quantity of incontinency episodes, so a CTM could greatly reduce the number of these episodes or the volume of the episodes but still not be reported as "successful" by our definition. The data collected were from the most recent clinic visit, therefore fluctuations in continence could also over- or underestimate reported continence rates [22]. Given that this study evaluated cross-sectional data, we were unable to determine the length of time that an individual had been on their CTM. There may have been individuals who recently changed to a new CTM and the effect of that change was not captured in the analysis.

One of the most striking findings of our analysis was the association of fecal continence with the socioeconomic factors of race/ethnicity and private insurance. Similar to the conclusions of previous studies that reported similar results, this finding is unlikely to be explained by biological factors alone [23]. This could be related to caregivers, providers, or both. Provider biases may exist in offering more invasive modalities to those of higher socioeconomic status or certain ethnic/racial groups. Some CTMs are not covered by all insurances within all states and therefore the ability to use a CTM may be affected by insurance coverage, socioeconomic status of the individual, and state of residence. Uncovered medical expenses and cost to families to attend multiple follow-ups for closely monitored CTMs may not be as easily attained by those of lower socioeconomic status. Antegrade enemas require surgery or percutaneous access, which creates a potential for variation in referral patterns.

It is clear from our analysis that there is not one overall best CTM for management of NBD. We think that the CTM hierarchy in Table 1 may be used as a guide for sequentially offering NBD treatments until continence is achieved. This suggestion follows updated expert panel guidelines for bowel function and care for people with SB which recommends beginning with non-intrusive treatments or dietary management ("Guidelines from the official Spina Bifida Website": https://www.spinabifidaassociation.org/guidelines/).

Sixty-percent of the continent individuals in this analysis appear to have achieved continence without surgical intervention. Antegrade enemas were associated with the highest continence rates in all age groups but require surgical intervention or percutaneous tube placement, often with revisions. In a meta-analysis of NBD management in SB, Velde et al. found that retrograde enemas resulted in a continence rate of 80%, compared to 81% for antegrade enemas, and 23% of the latter cohort required "redo" surgery for stoma complications [24]. Due to the invasive nature of antegrade enemas we believe they should not be offered without first determining if fecal continence can be achieved with a less invasive, non-surgical method.

Retrograde devices (transanal irrigation and cone/balloon large volume enemas) were associated with continence rates that were below that of antegrade enemas, but they were not greatly inferior and may be tried before surgical intervention.

The risks and benefits of any treatment modality must be weighed carefully and the choice of a CTM tailored for each individual and family. Sturkenboom et al. discussed the potential impact of antegrade enemas on outcomes such as pain, well-being, function, surgical complications, and quality of life, in addition to continence [25]. Optimization of a bowel treatment must include an evaluation of the need or want for independence and fecal continence, manual dexterity of the patient, ability of the patient to transfer to a commode or toilet for the program, and other factors that are unique to each individual.

The number of individuals able to complete their bowel management treatment independently were low for the entire group with only 23% of individuals able to carry out their regimen independently. The ability to complete a regimen independently increased

with age, as would be expected, to 33% in adults. Antegrade enemas and timed defecation were the CTMs most likely to be performed independently.

We did not analyze if starting interventions at a younger age would result in better continence rates because this was a cross-sectional and not longitudinal analysis. Hypothetically, earlier active management of NBD could result in better long-term outcomes by improving expectations in continence outcomes and possibly preventing long-term consequences of chronic constipation. This issue deserves further inquiry.

Proper and repeated education regarding the importance of NBD management and continence is crucial. Education should include a realistic expectation of continence and independence and an understanding that multiple CTMs likely will be trialed until a satisfactory regimen is found. Clinically, we believe that achieving continence in early childhood is advantageous to limit bullying, increase or preserve self-esteem, and improve quality of life associated with fecal incontinence [26–28].

The findings of our study must be interpreted in the context of several limitations. While definitions are provided for continence outcomes, these can be subject to variation in their interpretation and reporting. This raises particular caveats when attempting to compare outcomes among different centers, as the data may have been collected in subtly (though significantly) different ways. For example, continence data taken from face-to-face interviews may not be identical to that taken from written survey instruments, or data derived from clinician notes. Recall bias from individuals may also occur in reporting their continence. It is tempting to compare the effectiveness between the different CTMs, however a rigorous comparative analysis between CTMs was not performed. It was not carried out since this was cross-sectional and individuals utilizing a more invasive CTM may have previously had poorer results on less invasive therapy in the past, thus overestimating the latter's efficacy. Future analysis of longitudinal data in the NSBPR may better clarify that issue.

Similarly, selection bias may reduce the generalizability of these results. NSBPR involves multidisciplinary clinics at selected centers of excellence, so our findings may not be applicable to individuals with SB who either do not attend multidisciplinary SB clinics or attend clinics that are less rigorous than the clinics participating in the registry.

Despite these limitations, there are many strengths of this analysis. This is the first systematic analysis of a large cohort of individuals with MMC in regards to NBD and treatment outcomes. The analysis was carried out utilizing a novel hierarchical approach to the CTMs. Further studies assessing longitudinal data from this cohort would build on the effort to best manage these patients.

5. Conclusions

Our findings, in a large group of subjects with MMC and NBD, demonstrate statistically significant higher odds of continence in adolescents, females, and non-Hispanic whites, as well as those who have private insurance. Overall continence rates for those with NBD in this data registry group who used a CTM (or more than one) were modest, ranging from

36% to 69%. Although antegrade enemas were associated with the highest rate of continence in this population, our findings do not imply a causal connection between antegrade enemas and continence. Personalized treatment algorithms should be used for individuals with MMC to achieve continence, independence in carrying out their treatment, and satisfaction.

Supplementary Material

Refer to Web version on PubMed Central for supplementary material.

Acknowledgments

Abbreviations

CDC	Centers for Disease Control and Prevention
CTM	Criterion Treatment Modality
FC	Fecal continence
NBD	Neurogenic Bowel Dysfunction
NSBPR	National Spina Bifida Patient Registry
MMC	Myelomeningocele
SB	Spina Bifida

References

[1]. Krogh K, Christensen P, Sabroe S, Laurberg S. Neurogenic bowel dysfunction score. Spinal Cord. 2006; 44(10): 625–31. doi: 10.1038/sj.sc.3101887. [PubMed: 16344850]

[2]. Szymanski KM, Cain MP, Whittam B, Kaefer M, Rink RC, Misseri R. All incontinence is not created equal: impact of urinary and fecal incontinence on quality of life in adults with spina bifida. J Urol. 2017; 197(3 Pt 2): 885–91. doi: 10.1016/j.juro.2016.08.117. [PubMed: 28131501]

[3]. Wiener JS, Suson KD, Castillo J, Routh JC, Tanaka S, Liu T, et al. Bowel management and continence in adults with spina bifida: results from the national spina bifida patient registry 2009–15. J Pediatr Rehabil Med. 2017; 10(3–4): 335–43. doi: 10.3233/PRM-170466. [PubMed: 29125526]

[4]. Jinbo AK. The challenge of obtaining continence in a child with a neurogenic bowel disorder. J Wound Ostomy Continence Nurs. 2004; 31(6): 336–50. doi: 10.1097/00152192-200411000-00005. [PubMed: 15867709]

[5]. Liu CW, Huang CC, Yang YH, Chen SC, Weng MC, Huang MH. Relationship between neurogenic bowel dysfunction and health-related quality of life in persons with spinal cord injury. J Rehabil Med. 2009; 41(1): 35–40. doi: 10.2340/16501977-0277. [PubMed: 19197567]

[6]. Cunha TB, Tahan S, Soares MF, Lederman HM, Morais MB. Abdominal radiograph in the assessment of fecal impaction in children with functional constipation: comparing three scoring systems. J Pediatr (Rio J) 2012; 88(4): 317–22. doi: 10.2223/JPED.2199. [PubMed: 22915308]

[7]. Lynch AC, Antony A, Dobbs BR, Frizelle FA. Bowel dysfunction following spinal cord injury. Spinal Cord. 2001; 39(4): 193–203. doi: 10.1038/sj.sc.3101119. [PubMed: 11420734]

[8]. Lynch AC, Frizelle FA. Colorectal motility and defecation after spinal cord injury in humans. Prog Brain Res. 2006; 152: 335–43. doi: 10.1016/S0079-6123(05)52022-3. [PubMed: 16198711]

[9]. Stiens SA, Bergman SB, Goetz LL. Neurogenic bowel dysfunction after spinal cord injury: clinical evaluation and rehabilitative management. Arch Phys Med Rehabil. 1997; 78(3 Suppl): S86–102. doi: 10.1016/s0003-9993(97)90416-0. [PubMed: 9084372]

[10]. Powers CJ, George T, Fuchs HE. Constipation as a reversible cause of ventriculoperitoneal shunt failure. Report of two cases. J Neurosurg. 2006; 105(3 Suppl): 227–30. doi: 10.3171/ped.2006.105.3.227. [PubMed: 16970237]

[11]. Martinez-Lage JF, Martos-Tello JM, Ros-de-San Pedro J, Almagro MJ. Severe constipation: an under-appreciated cause of VP shunt malfunction: a case-based update. Childs Nerv Syst. 2008; 24(4): 431–5. doi: 10.1007/s00381-007-0514-3. [PubMed: 17926043]

[12]. Wide P, Mattsson GG, Drott P, Mattsson S. Independence does not come with the method-treatment of neurogenic bowel dysfunction in children with myelomeningocele. Acta Paediatr. 2014; 103(11): 1159–64. doi: 10.1111/apa.12756. [PubMed: 25048689]

[13]. Ojetti V, Bruno G, Paolucci V, Triarico S, D'Aversa F, Ausili E, et al. The prevalence of small intestinal bacterial overgrowth and methane production in patients with myelomeningocele and constipation. Spinal Cord. 2014; 52(1): 61–4. doi: 10.1038/sc.2013.131. [PubMed: 24247567]

[14]. McClurg D, Norton C. What is the best way to manage neurogenic bowel dysfunction? BMJ. 2016; 354: i3931. doi: 10.1136/bmj.i3931. [PubMed: 27815246]

[15]. Johnsen V, Skattebu E, Aamot-Andersen A, Thyberg M. Problematic aspects of faecal incontinence according to the experience of adults with spina bifida. J Rehabil Med. 2009; 41(7): 506–11. doi: 10.2340/16501977-0373. [PubMed: 19543660]

[16]. Choi EK, Im YJ, Han SW. Bowel management and quality of life in children with spina bifida in south korea. Gastroenterol Nurs. 2017; 40(3): 208–15. doi: 10.1097/SGA.0000000000000135. [PubMed: 26560901]

[17]. Le JT, Mukherjee S. Transition to adult care for patients with spina bifida. Phys Med Rehabil Clin N Am. 2015; 26(1): 29–38. doi: 10.1016/j.pmr.2014.09.007. [PubMed: 25479777]

[18]. Verhoef M, Lurvink M, Barf HA, Post MW, van Asbeck FW, Gooskens RH, et al. High prevalence of incontinence among young adults with spina bifida: description, prediction and problem perception. Spinal Cord. 2005; 43(6): 331–40. doi: 10.1038/sj.sc.3101705. [PubMed: 15685262]

[19]. Sawin KJ, Liu T, Ward E, Thibadeau J, Schechter MS, Soe MM, et al. The national spina bifida patient registry: profile of a large cohort of participants from the first 10 clinics. J Pediatr. 2015; 166(2): 444–50.e1. doi: 10.1016/j.jpeds.2014.09.039. [PubMed: 25444012]

[20]. Schechter MS, Liu T, Soe M, Swanson M, Ward E, Thibadeau J. Sociodemographic attributes and spina bifida outcomes. Pediatrics. 2015; 135(4): e957–64. doi: 10.1542/peds.2014-2576. [PubMed: 25780069]

[21]. Alabi NB, Thibadeau J, Wiener JS, Conklin MJ, Dias MS, Sawin KJ, et al. Surgeries and health outcomes among patients with spina bifida. Pediatrics. 2018 Sep; 142(3): e20173730. doi: 10.1542/peds.2017-3730. [PubMed: 30158199]

[22]. Liu T, Ouyang L, Thibadeau J, Wiener JS, Routh JC, Castillo H, et al. Longitudinal study of bladder continence in patients with spina bifida in the national spina bifida patient registry. J Urol. 2018; 199(3): 837–43. doi: 10.1016/j.juro.2017.11.048. [PubMed: 29132982]

[23]. Castillo J, Lupo PJ, Tu DD, Agopian AJ, Castillo H. The national spina bifida patient registry: a decade's journey. Birth Defects Res 2019 Aug 15; 111(14): 947–957. doi: 10.1002/bdr2.1407. [PubMed: 30403011]

[24]. Velde SV, Biervliet SV, Bruyne RD, Winckel MV. A systematic review on bowel management and the success rate of the various treatment modalities in spina bifida patients. Spinal Cord. 2013; 51(12): 873–81. doi: 10.1038/sc.2013.123. [PubMed: 24126852]

[25]. Sturkenboom R, van der Wilt AA, van Kuijk SMJ, Ahmad A, Janssen PT, Stassen LPS, et al. Long-term outcomes of a Malone antegrade continence enema (MACE) for the treatment of fecal incontinence or constipation in adults. Int J Colorectal Dis. 2018; 33(10): 1341–1348. doi: 10.1007/s00384-018-3088-5. [PubMed: 29934702]

[26]. Roach MJ, Frost FS, Creasey G. Social and personal consequences of acquired bowel dysfunction for persons with spinal cord injury. J Spinal Cord Med. 2000; 23(4): 263–9. doi: 10.1080/10790268.2000.11753535. [PubMed: 17536296]

[27]. Choi EK, Shin SH, Im YJ, Kim MJ, Han SW. The effects of transanal irrigation as a stepwise bowel management program on the quality of life of children with spina bifida and their caregivers. Spinal Cord. 2013; 51(5): 384–8. doi: 10.1038/sc.2013.8. [PubMed: 23439065]

[28]. Marte A, Borrelli M. Transanal irrigation and intestinal transit time in children with myelomeningocele. Minerva Pediatr 2013; 65(3): 287–93. [PubMed: 23685380]
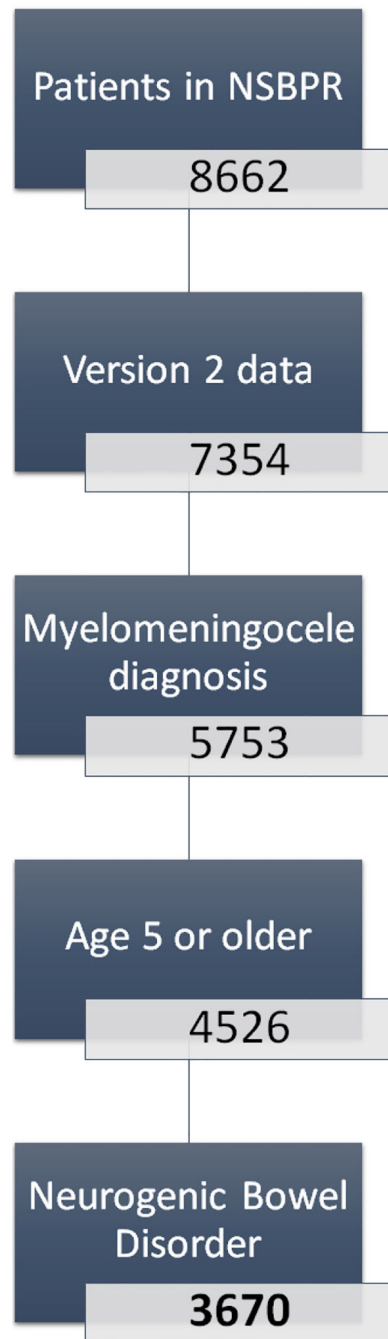
Fig. 1.
CONSORT of study population from National Spina Bifida Patient Registry, December 2017. Version 2 data collection started October 2013; additional questions were added to the registry to obtain more detailed data on outcomes, additional procedures, imaging studies, and lab results. Also, two other diagnoses (Terminal Myelocystocele and Split Cord Malformation) were added to the eligible diagnoses. Specifically, answer choices for continence were changed from previous Yes/No to more detailed incontinence frequency.

Table 1

Criterion treatment modalities included in National Spina Bifida Patient Registry in order of invasiveness (least to most)

No intervention
Timed defecation
Oral medications
Digital rectal stimulation
Suppositories
Mini-enemas
Standard rectal enemas
Manual disimpaction
Cone/balloon large volume enema
Peristeen® transanal irrigation system
Antegrade enemas (MACE, Chait, Other)
Pouched fecal diversion (colostomy/ileostomy)

Table 2

Key demographic and clinical characteristics of individuals ≥ 5 years of age with myelomeningocele and neurogenic bowel dysfunction in the National Spina Bifida Patient Registry, November 2013–December 2017

Variables	Overall n (%) N = 3670	N (%) or statistics by age at last visit				
		5–11 n = 1445	12–19 n = 1240	20 and older n = 985	P-value[+]
Sex						
Male	1782 (48.6)	731 (50.6)	620 (50.0)	431 (43.8)	0.002
Female	1888 (51.4)	714 (49.4)	620 (50.0)	554 (56.2)	
Race/ethnicity (N = 3653)						
Non-Hispanic White	2301 (63.0)	803 (56.0)	716 (57.9)	782 (79.5)	< 0.001
Non-Hispanic Black	291 (8.0)	104 (7.3)	112 (9.1)	75 (7.6)	
Hispanic or Latino	831 (22.7)	387 (27.0)	351 (28.4)	93 (9.5)	
Other	230 (6.3)	139 (9.7)	57 (4.6)	34 (3.5)	
Level of motor function						
Thoracic	799 (21.8)	163 (11.3)	247 (19.9)	389 (39.5)	< 0.001
High-Lumbar	457 (12.5)	167 (11.6)	163 (13.1)	127 (12.9)	
Mid-Lumbar	1161 (31.6)	500 (34.6)	399 (32.2)	262 (26.6)	
Low-Lumbar	622 (16.9)	306 (21.2)	213 (17.2)	103 (10.5)	
Sacral	631 (17.2)	309 (21.4)	218 (17.6)	104 (10.6)	
Health insurance						
Any private	1574 (42.9)	631 (43.7)	577 (46.5)	366 (37.2)	< 0.001
Non-private	2096 (57.1)	814 (56.3)	663 (53.5)	619 (62.8)	< 0.001

*
Statistically significant at p ≤ 0.05 level.

[+]
Chi-square test.

Table 3

Prevalence of fecal continence ** by criterion treatment modality and age in individuals with myelomeningocele and neurogenic bowel dysfunction in the National Spina Bifida Patient Registry, November 2013–December 2017

Variables	Overall N (%) (N =3670)	n (%) within each CTM who were continent*	n (%) within each age group who were continent			
			5–11 (n = 1445)	12–19 (n = 1240)	20 and older (n = 985)	P-value[+]
Criterion treatment modality						
No intervention	519 (14.1)	0 (0)				
Timed defecation	122 (3.3)	56 (45.9)	11 (22.4)	30 (55.6)	15 (78.9)	<0.001
Oral medications only	828 (22.6)	329 (39.7)	71 (22.7)	128 (48.9)	130 (51.4)	<0.001
Digital rectal stimulation	117 (3.2)	63 (53.8)	9 (39.1)	12 (42.9)	42 (63.6)	0.052
Suppositories	214 (5.8)	88 (41.1)	38 (34.5)	35 (47.9)	15 (48.4)	0.132
Mini-enemas	168 (4.6)	83 (49.4)	49 (46.2)	30 (55.6)	4 (50.0)[++]	0.55
Standard rectal enemas	128 (3.5)	72 (56.3)	20 (37.0)	18 (62.1)	34 (75.6)	<0.001
Manual disimpaction	92 (2.5)	33 (35.9)	6 (18.8)	9 (30.0)	18 (60.0)	0.002
Cone/balloon large volume enemas	371 (10.1)	217 (58.5)	133 (56.6)	49 (66.2)	35 (56.5)	0.32
Peristeen® transanal irrigation	76 (2.1)	44 (57.9)	21 (45.7)	20 (80.0)	3 (60.0)[++]	0.013
Antegrade enemas	971 (26.5)	666 (68.6)	169 (66.0)	319 (69.7)	178 (69.3)	0.58
Pouched fecal diversion	64 (1.7)					

[+] Chi-square test.

[++] Exact test was performed when expected cell count was 5 or less.

[*] Overall comparison of continence rate in each modality group, p < 0.0001.

[**] Fecal continence is defined as having episodes of incontinence less than once a month or never.

Table 4

Summary of multiple logistic regression on fecal continence[+] among individuals $\geq$ 5 years of age with myelomeningocele and neurogenic bowel dysfunction in the National Spina Bifida Patient Registry, November 2013–December 2017 ($n = 3589$, patients with pouched fecal diversion were excluded)

Variables	Odds ratio (95% CI)	P-value
Age group		< 0.001[‡]
5 to < 12[†]		
12 to < 20	1.99 (1.70–2.34)	< 0.001
20 or older	1.82 (1.51–2.18)	< 0.001
Gender		
Male[†]		
Female	1.24 (1.08–1.42)	0.002
Race/ethnicity (17 missing)		< 0.002[‡]
Non-Hispanic White[†]		
Non-Hispanic Black	0.61 (0.47–0.79)	< 0.001
Hispanic or Latino	0.73 (0.61–0.87)	< 0.001
Other	0.71 (0.53–0.95)	0.021
Functional level of motor function		0.008[‡]
Thoracic[†]		
High-Lumbar	0.98 (0.77–1.25)	0.87
Mid-Lumbar	1.09 (0.90–1.33)	0.37
Low-Lumbar	1.37 (1.09–1.72)	0.006
Sacral	1.34 (1.07–1.68)	0.011
Insurance		
Any private[†]		
Non-private	0.61 (0.53–0.70)	< 0.001
Oral agent use		
Yes[†]		
No	1.16 (1.01–1.33)	0.031

[†]Reference group.

[‡]Overall *p*-value for variables with more than 2 categories.

[+]Fecal continence is defined as having episodes of incontinence less than once a month or never.

Table 5

Prevalence of independent use by criterion treatment modality and age in individuals with myelomeningocele and neurogenic bowel dysfunction in the National Spina Bifida Patient Registry, November 2013–December 2017

Variables	Overall *n* (%) (N = 3670)	N independent (%)	*n* independent (%)				
			5–11 (*n* = 1445)	12–19 (*n* = 1240)	20 and older (*n* = 985)	*P*-value[+]
Criterion treatment modality							
No intervention	519 (14.1)	–	–	–	–	
Timed defecation	122 (3.3)	68 (55.7)	15 (30.6)	38 (70.4)	15 (78.9)	< 0.001
Oral medications only	828 (22.6)	–	–	–	–	
Digital rectal stimulation	117 (3.2)	47 (40.2)	# (< 15.0)[**]	10 (35.7)	34 (51.5)	0.003
Suppositories	214 (5.8)	49 (22.9)	6 (5.5)	25 (34.2)	18 (58.1)	< 0.001
Mini-enemas	168 (4.6)	30 (17.9)	7 (6.6)	21 (38.9)	# (< 30.0)[**]	< 0.001
Standard rectal enemas	128 (3.5)	33 (25.8)	# (< 10.0)[**]	11 (37.9)	17 (37.8)	< 0.001
Manual disimpaction	92 (2.5)	22 (23.9)	# (< 15.0)[**]	9 (30.0)	9 (30.0)	0.181
Cone/balloon large volume enemas	371 (10.1)	78 (21.0)	23 (9.8)	27 (36.5)	28 (45.2)	< 0.001
Peristeen® transanal irrigation	76 (2.1)	10 (13.2)	# (< 5.0)[**]	# (< 25.0)[**]	# (< 65.0)[**]	0.003
Antegrade enemas	971 (26.5)	478 (49.2)	56 (21.9)	246 (53.7)	176 (68.5)	< 0.001
Pouched fecal diversion	64 (1.7)	34 (63.0)[a]	# (< 30.0)[**]	# (< 60.0)[**]	27 (71.1)	0.097

[+] Chi-square test.

[**] Exact test was performed when expected cell count was 5 or less; cell count of 5 or less not shown (#).

[a] There were 10 individuals with missing data.

Table 6

Prevalence of fecal continence [++] by criterion treatment modality and use of any oral agent in individuals ≥ 5 years of age with myelomeningocele and neurogenic bowel dysfunction in the National Spina Bifida Patient Registry, November 2013–December 2017

Variables	Overall n (%) (N = 3087[+++])	n continent (%)		P-value[+]
		With oral agents	Without oral agents	
Criterion treatment modality				
Oral medications only	828 (22.6)	329 (39.7)	–	
Timed defecation	122 (3.3)	28 (37.8)	28 (58.3)	0.040
Digital rectal stimulation	117 (3.2)	16 (31.4)	47 (71.2)	< 0.001
Suppositories	214 (5.8)	45 (34.6)	43 (51.2)	0.023
Mini-enemas	168 (4.6)	54 (46.6)	29 (55.8)	0.32
Standard rectal enemas	128 (3.5)	20 (39.2)	52 (67.5)	0.002
Manual disimpaction	92 (2.5)	9 (17.6)	24 (58.5)	< 0.001
Cone/balloon large volume enemas	371 (10.1)	71 (54.6)	146 (60.6)	0.27
Peristeen® transanal irrigation	76 (2.1)	13 (46.4)	31 (64.6)	0.152
Antegrade enemas	971 (26.5)	138 (60.8)	528 (71.0)	0.00423

[+] Chi-square test.

[++] Fecal continence is defined as having episodes of incontinence less than once a month or never.

[+++] Those using no intervention (n = 519) or pouched fecal diversions (n = 64) were excluded from analysis.

J Pediatr Rehabil Med. Author manuscript; available in PMC 2022 January 20.