



Reliability and Validity of the Neurogenic Bladder Symptom Score in Adults with Cerebral Palsy

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OBJECTIVE	To examine the utility of the Neurogenic Bladder Symptom Score (NBSS) in adults with cerebral palsy (CP).
METHODS	Patients participated in a structured intake which included bladder management. Patients (or caregivers, if patients unable) completed NBSS and SF-Qualiveen twice over a 2-week period. Validity was determined using Cronbach's alpha and correlation testing between NBSS, intake, and SF-Qualiveen. Reliability was determined using test-retest method and intraclass correlation coefficients.
RESULTS	Fifty-four patients were included. Thirty-six patients (67%) used a wheelchair; 35 patients (66%) required a caregiver to complete questionnaires. Median NBSS subdomain scores were 12 of 29 for incontinence, 9 of 22 for storage and/or voiding, 2 of 23 for consequences and 1 of 4 for quality of life (QOL). Lower scores reflect fewer symptoms. Reliability was high (intraclass correlation coefficients = 0.90). There was a moderate correlation ($R = 0.70$) between NBSS QOL and SF-Qualiveen. Bladder management method, as determined by the NBSS, was indwelling catheter in 4, intermittent catheterization in 6, voiding into a toilet in 33, and missing in 11. Importantly, there was no option for voiding into a diaper, which was common in this population.
CONCLUSION	For adults with CP, the NBSS has high reliability and statistically, it demonstrated appropriate validity, but it has limitations. Its face validity is questionable given that diapers were not an option. The validity of caregiver completion needs further assessment. The NBSS may have a floor effect for detecting urinary consequences or QOL, reflected by consistently low scores in these subdomains. The development of a specific urinary symptom and/or QOL tool for adults with CP is needed. UROLOGY 128: 107–111, 2019. © 2019 Elsevier Inc.

Cerebral palsy (CP) is a nonprogressive, but often changing motor impairment secondary to brain injury arising in early development. It is the most common motor disability in childhood. The prevalence is roughly 3.1-3.6 for every 1000 children.¹ Etiologies include prematurity, fetal hypoxia, and maternal infection during gestation.²⁻⁴ Urinary incontinence and voiding dysfunction are common in children with CP, but studies are somewhat lacking.⁵⁻⁷ Even less is known about the urologic management of adults with CP. We previously demonstrated that half of adult patients seen at our CP referral center had concerning findings on urodynamics,

highlighting the need for urologic assessment.⁸ However, the urinary symptoms of adults with CP have not been well-described using any validated assessment tool.

The Neurogenic Bladder Symptom Score (NBSS) is a 24-question instrument used to measure the symptoms of either acquired or congenital neurogenic bladder (NGB). The first question classifies patients by primary bladder management method (indwelling catheter and/or urostomy, condom catheter, intermittent catheterization, or toileting). The next 22 questions cover 3 domains: incontinence, voiding, and consequences. A final question addresses overall quality of life (QOL).⁹ It has been validated for use in adults with spinal cord injury (SCI), spina bifida, and multiple sclerosis (MS).^{9,10} However, it has not been studied for use in patients with CP.

The severe motor spasticity characteristic of CP creates unique voiding and communication challenges. Spasticity results in pseudo-dyssynergia and difficulty opening the legs, both of which complicate catheterization and toileting. Spasticity and cognitive dysfunction both lead to verbal and written communication challenges that might

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preclude use of standard questionnaires. Given the unique characteristics of CP and the lack of any currently accepted method of urinary symptom assessment in this population, we sought to determine the validity and reliability of the NBSS in adults with CP.

MATERIALS AND METHODS

After Institutional Review Board approval, adult patients with CP were recruited from 2 transitional urology clinics in the United States. We excluded patients who were expected to have acute changes in their bladder function (for example due to a new medication or a current urinary infection).

During the first visit, patients completed a structured intake interview, which included demographic details along with general questions about the severity of their CP and their primary bladder management. They also completed: (1) the NBSS, (2) a global bladder function question (how much of a problem is your bladder and/or urinary function, 1 (no problem) 10 (very significant problem), and (3) the SF-Qualiveen, which is a validated instrument for measuring urinary difficulties in people with SCI or multiple sclerosis.¹¹ The SF-Qualiveen focuses on how bladder management affects QOL; it includes questions describing worry about bladder management, embarrassment or having to regulate life activities because of one's bladder issues. It has been assessed to have a readability of roughly an eighth grade level.¹² These questionnaires have been validated in English language. If patients were unable to complete the questionnaires independently due to any limitations, then we allowed caregivers to complete the study questions for them. We acknowledge that the validity of caregiver completion of these 2 questionnaires has not been previously assessed, and the psychometric properties can vary based on who completes the instrument.¹³ At visit 2 (approximately 2 weeks later), patients and/or caregivers were asked to complete the NBSS a second time.

Validity Assessment

First, we compared the bladder management as determined from the intake interview to the answers to the NBSS question 1 (primary bladder management method). We considered the former to be the gold standard due to the ability to ask clarifying follow-up questions and allowing more than one method of bladder

management. Construct validity was determined by testing hypothesized correlations between the NBSS total score or NBSS domain scores and the global bladder function or SF-Qualiveen.

Reliability Assessment

Cronbach's alpha was used to measure internal consistency within a particular NBSS domain. A value >0.70 is considered a good indicator of internal validity. Test-retest reliability of the NBSS and its domains was calculated using the visit 1 and visit 2 NBSS scores.

In order to assess the potential impact of caregiver completion, we repeat the validity and reliability analysis with stratification of patients who completed the questionnaires themselves compared to those who had a caregiver complete them. While our study was not powered for this comparison, it provided a unique opportunity for hypothesis-generating insights.

Statistical Analysis

Data is summarized as proportions and medians (interquartile ranges). Patients with more than one question missing from an NBSS domain were excluded from the relevant analysis (n = 14); imputation was used to account for missing data when only one question was missing in a domain. Our sample size was selected based on the hypothesis testing approach to sample size calculation for reliability; we assumed an alpha = 0.05, beta = 0.20; with our minimum Cronbachs alpha set at 0.80, and our expected Cronbachs alpha set at 0.90, yielding a minimum of 46 people.¹⁴ Differences in proportions were determined using a Fisher exact test. The Pearson correlation coefficient was used to assess construct validity correlations. We considered an r > 0.7 as a strong relationship, r = 0.7 to 0.3 as moderate, and r < 0.3 as a weak correlation. An intraclass correlation coefficient (ICC 2,1) was used to assess agreement for the test-retest reliability assessment. SPSS version 24 was used for all calculations, and a P < .05 was considered significant.

RESULTS

In total, 54 patients were included for analysis (Table 1). The median age was 30 years (interquartile range 25-38), approximately half were male, and the majority required a wheelchair

Table 1. Baseline characteristics

	Total Cohort	Independently Completed (n = 18)	Caregiver Completed (n = 35)
Median Age	30 (25-38)	38 (27-49)	29 (24-34)
Male	29 (55%)	8 (44%)	21 (62%)
Median BMI (kg/m ²)	23 (20-27)	27 (23-31)	21 (20-25)
Employed?			
No	35 (66%)	8 (44%)	26 (76%)
Yes	16 (30%)	9 (50%)	7 (21%)
Student/Other	2 (4%)	1 (6%)	1 (3%)
Mobility			
Wheelchair	36 (67%)	7 (39%)	28 (80%)
Cane/Walker	12 (22%)	6 (33%)	6 (17%)
Stand with help	4 (7%)	4 (22%)	0 (0%)
Walk sometime or all the time	2 (4%)	1 (6%)	1 (3%)
Chronic Pain	20 (37%)	10 (56%)	9 (26%)
Spasticity	51 (94%)	16 (89%)	34 (97%)

Table 2. Structured interview responses and NBSS domain scores in patients with and without caregivers completing the questionnaires

	Total	Independently Completed (n = 18)	Caregiver Completed (n = 35)
Interview questions, n (%)			
Void to toilet*	27 (51%)	14 (82%)	12 (34%)
Pads/diapers*	28 (52%)	5 (28%)	23 (66%)
NBSS domains (Median, IQR)			
Incontinence (Max 29)	12 (3-17)	10 (2-14)	15 (8-18)
Storage/void (Max 22)	9 (6-12)	11 (5-14)	9 (7-12)
Consequences (Max 23)	2 (0-5)	3 (0-5)	1 (0-5)
QOL (Max 4)	1 (0-2)	1 (0-2)	1 (0-2)
NBSS total (Max 78)	24 (10-32)	21 (9-33)	26 (13-32)

* Some patients report using both methods, therefore % do not necessarily sum to 100%.

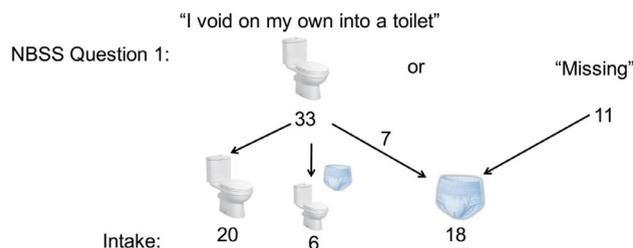


Figure 1. Interpretation of NBSS question 1 using structured interview answer. (Color version available online.)

and reported spasticity (67% and 94%, respectively). Thirty-five patients (66%) had a caregiver complete the questionnaires for them.

The results of the NBSS for the first visit are shown in Table 2. In general, there were minimal missing data, with imputation required for only 1.4% of the question responses. Bladder management (NBSS question 1) was reported as “indwelling and/or urostomy” in 4 patients (9%), “Clean intermittent catheterization” in 6 patients (11%), and “voiding” in 33 patients (61%). No one reported using “condom catheter,” and the answer was left blank for the remaining 11 patients (20%). These answers from the NBSS were then compared to the structured interview questions (Fig. 1). Of the 33 patients who reported voiding into a toilet on the NBSS, only 20 reported voiding to a toilet as their only method of voiding in the structured interview. Six of the 33 were using a combination of diapers and toileting, while 7 were actually completely diaper dependent. All 11 patients with a

missing answer to NBSS question 1 were also diaper dependent. Compared to those who did not require a caregiver, patients requiring caregiver assistance were significantly more likely to utilize pads and/or diapers and significantly less likely to void into a toilet ($P < .01$). Regarding NBSS domain scores, a lower score represents fewer symptoms. The median scores for NBSS Incontinence and storage and/or voiding had appropriate distributions, with no ceiling effects and a minor floor effect. The consequences domain had low scores with floor effects, and the QOL question also tended to have floor effects. NBSS Storage/Voiding and QOL scores were similar between patients who required a caregiver compared to those who did not (Table 2). However, the NBSS Incontinence domain was slightly higher for patients requiring a caregiver (15 vs 10, $P = .05$), implying more incontinence in patients with a caregiver. Patients with a caregiver were less likely to report chronic pain than those with a caregiver (26% vs 56%, $P = .04$).

There was good internal consistency for the NBSS domains of incontinence, storage/voiding, and the total score (Cronbach’s alpha of 0.90, 0.76, and 0.89, respectively); however, the consequences domain had a Cronbach’s alpha of only 0.59. The hypothesized relationships and the actual relationships between the NBSS domains and the global bladder function question and SF-Qualiveen are shown in Table 3. The majority of the relationships were consistent with the direction and strength of our hypotheses, and the three that were not actually showed stronger than expected relationships.

The ICC for test-retest reliability was high for the NBSS domains incontinence (0.84; 0.71-0.92), storage and/or voiding (0.90; 0.82-0.95), consequences (0.81; 0.67-0.89), the QOL question (0.76; 0.60-0.87) and the total score (0.90; 0.82-0.95).

Table 3. Correlation between NBSS domain Scores and SF-Qualiveen and global bladder function question. All correlations have a $P < .01$.

	Incontinence	Storage/Voiding	Consequences	QOL	Total
SF-Qualiveen					
Hypothesis	+Moderate	+Weak	+Weak	+Moderate	+Moderate
Actual	0.50	0.47	0.62	0.70	0.64
Result	✓	X	X	✓	✓
Global bladder function					
Hypothesis	+Moderate	+Weak	+Moderate	+Strong	+Strong
Actual	0.53	0.57	0.60	0.74	0.71
Result	✓	X	✓	✓	✓

DISCUSSION

The search for valid and reliable assessment tools of urinary symptoms and QOL remains a challenge in patients with neurogenic bladder. There are numerous questionnaires available but the literature provides little guidance as to which to use for the different phenotypes of NGB (eg, overactive vs underactive bladder, on clean intermittent catheterization or not, or NGB due to multiple sclerosis vs Parkinson's disease vs CP).^{15,16} The NBSS represents one validated questionnaire for SCI, spina bifida, and MS. Herein we demonstrate that the NBSS performs well in adults with CP and that with some minor modifications, it could prove to be a valuable tool in this unique patient population.

When working to identify a questionnaire to use in a clinical setting, such as the assessment of symptoms in NGB secondary to CP, it is preferable to use existing questionnaires. If existing questionnaires can be validated in the population of interest, then the time and expense involved in developing a new assessment tool can be avoided. Furthermore, use of an existing tool allows different populations to be compared with each other using the same tool. However, some disease phenotypes may be unique enough that they require their own assessment tool. Even though CP is a suprapontine injury and exhibits detrusor overactivity just like other cortical level conditions such as stroke, the skeletal muscle spasticity characteristic of CP manifests as a unique phenotype of NGB. Pelvic floor spasticity results in outlet obstruction and resultant chronic urinary retention in some due to high external sphincter tone during voiding (termed pseudo-dyssynergia). Furthermore, the lower extremity spasticity and difficulty catheterizing through the tight external sphincter make it challenging to employ urethral intermittent catheterization. This unique bladder phenotype combined with communication challenges and dependence on a caregiver made us question whether the NBSS was valid for use in CP.

Overall, the NBSS performed well for assessment of urinary symptoms in adults with CP. The test-retest reliability and internal consistency were both high. Further, the scores on the NBSS correlated well with the global assessment of bladder function and the Qualiveen-SF, suggesting that content validity was good.

However, our study highlights some difficulties in using the NBSS in CP. Notably, 24 of our patients (44%) either void into a diaper as a primary bladder management or use a toilet some time and diaper the rest of the day. Yet the NBSS bladder management question does not include a response for voiding to a diaper. Fortunately, this question is a nonscored item and could easily be modified for people with CP, without affecting the measurement properties of the NBSS.

Communication and mobility challenges are common for adults with moderate to severe CP, and many with advanced CP rely on caregivers for activities of daily living. In a study from Belgium, those most severely affected

by CP were far more likely to experience incontinence.¹⁷ In our cohort, 66% of patients required a caregiver to complete the questionnaires, and we noted some differences in the NBSS based on this variable. Of note, the NBSS Incontinence score was higher in patients who had a caregiver complete the questionnaires, suggesting worse incontinence in those more severely affected by CP. Alternatively, patients without a caregiver may have under-reported their level of incontinence. When a caregiver completed the questionnaire the reported incidence of chronic pain was lower (26% vs 56%); as it is unlikely that people with more severe CP have less pain, this begs the question of whether caregivers under-report pain experienced by their dependents with CP. Overall, the NBSS Consequences and QOL domains were scored very low for nearly all patients. Although not significant, the consequences actually trended lower (better) in patients who had a caregiver complete the questionnaire. Like the pain question, this may mean that patients with a caregiver truly had low rate of "consequences," such as infection, stones, or medications or, more likely, that caregivers for adults with CP may not be able to discern these consequences without adequate communication with the patient, thus resulting in a "floor effect" for the use of NBSS in patients with the most severe CP. The validity of caregiver completion requires further assessment.

Limitations of our study include the utilization of only 2 sites for patient recruitment. While we did attempt to include patients with a spectrum of CP, we recruited patients from adult urology clinics. Therefore, our cohort is likely over-represented by people with more severe CP. Therefore, our findings may not be applicable to adults with mild to moderate CP or children. While we attempted to utilize a structured patient intake as well as alternative questionnaires, there was no "gold standard" for comparison to the NBSS. Another limitation was the challenge of interpreting data from a questionnaire obtained with caregiver assistance. While we performed an assessment of the severity of CP in terms of spasticity and pain, we did not assess communication limitations. Thus, caregiver involvement was likely for a variety of reasons—both lack of dexterity and verbal limitations. Future research efforts regarding tools to assess bladder specific QOL for adults with CP could be more adaptive (given frequent communication challenges) and could include the management strategy voiding into diaper spontaneously. Adaptations could include a touch screen or eye-tracking software for symptom assessment. We suspect that structured patient and caregiver interviews may elucidate other relevant interview questions for this unique patient population.

CONCLUSION

The NBSS has high reliability and statistically appropriate validity. However, it has limitations in its current state

when assessing adults with CP. Its face validity is questionable given the additional method of voiding in diapers, which was not considered when developing the NBSS. Further assessment of validity is needed, especially regarding how responses may change when a caregiver needs to fill out the questionnaire for the patient. Adaptive technology that allows the patient rather than the caregiver to complete the questionnaire may address this issue. With modifications and adaptive technology, the NBSS may serve as a useful tool for assessing urinary symptom and QOL in adults with CP.

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