Bladder Bowel Dysfunction Scoring System

A novel, illustrated questionnaire for evaluation of voiding dysfunction in children

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ABSTRACT

INTRODUCTION: We sought to evaluate the reliability and validity of a new, illustrated questionnaire, the Bladder Bowel Dysfunction Scoring System (BBDSS) in the assessment of overactive bladder (OAB) and bladder bowel dysfunction (BBD).

METHODS: The BBDSS questionnaire consisted of 12 structured questions. This pilot study was designed with two principal groups of questions: one group to assess bladder symptoms and the other to assess bowel dysfunction during the preceding month. Each question had three possible answers, with each answer being assigned a severity score. We prospectively collected previously untreated patients referred to our voiding dysfunction clinic for the first time. A control group of healthy children was recruited to assess the reliability of the BBDSS questionnaire. The provisional diagnosis was collected from patients' charts at the time of presentation.

RESULTS: The questionnaire was administered to 92 children (44 in the affected group and 48 in the control group). The age at presentation was similar in both groups (17 months or nine years, nine months). The mean total score for the affected group was 8.7 (3–14), while it was 1.19 (0–5) for the control group ($p<0.001$). There was a strong correlation, between the total BBDSS score and both groups ($r=0.88$, $p<0.001$). Using the receiver operating characteristic (ROC) curve, the BBDSS was found to be an excellent tool in differentiating normal from affected patients (area under the curve [AUC]=0.98, $p<0.001$). When the total BBDSS score was ≥6, the positive predictive value was 1, with a negative predictive value of 0.89. The defecation part of the BBDSS was a good tool in differentiating OAB from BBD patients (AUC=0.89, $p<0.001$). No patient with OAB had a bowel score >3.

CONCLUSIONS: The BBDSS is a reliable and valid instrument in the diagnosis of voiding dysfunction. The questionnaire was easily administered by parents or children. Moreover, it can differentiate between OAB and BBD.

INTRODUCTION

Voiding dysfunction is a common problem in childhood. Constipation is considered a cause or a common association with bladder dysfunction in 50% of cases. One-third of children with constipation treated with laxatives will have their voiding issues resolved.

Most published questionnaires in the literature evaluate constipation by one, or at most a few questions that may be considered vague for many parents.[1-4] Hence, poor information about the child's bowel habits could lead to inappropriate management of the underlying problem. Moreover, some of the available questionnaires are long, time-consuming, and even considered boring to be answered by some parents and children. This may hamper the effectiveness of the questionnaire to fully delineate the scope of the problem.

The need for a new questionnaire is warranted to overcome the above-mentioned disadvantages. We designed the Bladder Bowel Dysfunction Score System (BBDSS) to evaluate both bladder and bowel dysfunction, keeping in mind the inherent limitations of previous questionnaires. To our knowledge, this is the first illustrated questionnaire that is designed to differentiate between overactive bladder (OAB) and bladder bowel dysfunction (BBD).

Our aim was to conduct a pilot study to assess the reliability and validity of the BBDSS for the evaluation of voiding dysfunction. Furthermore, we wished to assess the cutoff point for the diagnosis of bladder and/or bowel dysfunction.

METHODS

We developed a new, self-administrated questionnaire with illustrations, BBDSS, to aid in the diagnosis and management of voiding dysfunction in children. The BBDSS questionnaire consisted of 12 structured questions. The questionnaire was designed with two principal groups of questions: one group to assess bladder symptoms and the other to assess bowel dysfunction during the preceding month. Bladder symptoms included incontinence (day and night), frequency, quality of the urinary stream, urgency, and holding maneuvers. Defecation evaluation questions were based on the ROME IV criteria, including frequency, stool consistency, encopresis, painful bowel movements, and presence of large diameter stools that could block the toilet.[5] We included diagrams illustrating various types of stool consistency experienced during bowel movements. These diagrams were illustrative of hard stools (types 1, 2, and 3) and normal stool (type 4), as defined by the Bristol stool form scale.[6] Moreover, we added illustrations for normal and abnormal urinary streams. Normal urinary stream was defined as a continuous strong flow of urine. Abnormal urinary stream was defined as thin, interrupted, and/or urinary spraying.

Every question had three possible answers: 1) no; 2) half of the time; or 3) most of the time. Each answer was assigned a specific score related to the severity of the parameter assessed, i.e., if the answer was "no," the assigned severity score was "0" and if it was "most of the time," the assigned severity score was "2." Finally, for each completed questionnaire the scores were summed for the bowel and bladder questions separately (Figure1).

After obtaining local ethical approval (#2017-3332), we prospectively collected patients who were referred to our voiding dysfunction clinic for the first time, from March 2018 to June 2020. Patients who were on medication at the time of presentation were excluded from the study. Moreover, we excluded patients with neurogenic bladder or those having urologic anomalies including phimosis, stone disease, hypospadias, and hydronephrosis. The target age was 6–14 years.

The BBDSS questionnaire was initially pretested on five patients to ensure that all questions and illustrations were clearly understood and then we subsequently began recruiting patients who met our inclusion criteria. Before answering the questionnaire, parental consent was obtained after informing them of the content and the study objectives of the questionnaire. Despite the questionnaire having been designed as a self-administrated instrument, we freely offered support should problems be encountered by the parents or patients.

The attending physicians were blinded to the questionnaire answered by patients. We collected the working diagnosis from patients' charts at the time of presentation. The diagnosis was considered as primary voiding dysfunction (no associated constipation) and BBD syndrome (associated constipation).

Another group of healthy children of the same age was recruited to assess the reliability of the BBDSS questionnaire (control group). Patients were collected from otolaryngology and ophthalmology clinics. We confirmed that all patients in the control group children were free of bladder or bowel problems at the time of administering the questionnaire by verbal confirmation with the parents and patients.

We defined OAB, according to the International Children's Continence Society, as urinary urgency, which is usually accompanied by frequency and nocturia, with or without urinary incontinence, after the exclusion of urinary tract infection and other contributing pathologies.[7] Moreover, the diagnosis of BBD is considered when a patient has both voiding and bowel dysfunctions.

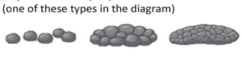
The primary outcome of this pilot study was to evaluate the validity and reliability of the BBDSS. The second outcome was to test the association between the BBDSS and the primary diagnosis at the time of the questionnaire (either OAB or BBD).

BLADDER-BOWEL DYSFUNCTION SCORE SYSTEM

➢ **Bladder issue evaluation**

During the **last month**	NO	SOMETIMES (1-3 days/week)	MOST OR ALL THE TIME (≥ 4days/week)
Do you wet your underwear during the day?	0	1	2
Do you wet underwear during sleep?	0	1	2
Do you rush to the washroom to pee?	0	1	2
Is the urine stream **abnormal ?** 	0	1	2
Does it hurt or burn when you pee?	0	1	2
Do you pee **more than 7 times everyday**?	0	1	2
Do you hold your pee by crossing legs, squatting, or doing the "pee dance"?	0	1	2

➢ **Bowel issue evaluation**

During the **last month**	NO	SOMETIMES (≤ half month)	MOST OR ALL THE TIME (> half month)
Do you have hard poop? (one of these types in the diagram) 	0 (*Normal like this diagram*) 	1	2
Do you poop **less than 3 times /week**?	0	1	2
Do you soil your underwear **at least once a week**?	0	1	2
Is your poop painful?	0	1	2
Does your poop block the toilet?	0	1	2

A question related to the Questionnaire (during the study only)

Are words or sentences understandable and clear?

NO YES ALMOST (If applicable, please mark the unclear question)

Figure 1. The Bladder Bowel Dysfunction Scoring System.

BBDSS questionnaire evaluation

SPSS 20® was used for data collection and statistical analysis. The BBDSS questionnaire underwent multiple steps to be evaluated as a valid instrument for voiding dysfunction diagnosis. First, the reliability of the questionnaire was evaluated using the Mann-Whitney test and the Pearson product-moment correlation coefficient to compare the scores of both groups. Thereafter, the BBDSS validity was tested in comparison to the collected diagnoses using the Mann-Whitney test. To determine the accuracy of the questionnaire in classifying the children (affected vs. control), the discrimination function analysis was used. The internal consistency of the questionnaire was evaluated using Cronbach's alpha test. A receiver operating characteristic (ROC) curve was used to assess the cutoff point related to the diagnosis of bladder and/or bowel dysfunction. Lastly, we correlated the initial diagnosis with the BBDSS score. The test is statistically significant if $p<0.05$.

RESULTS

Inclusions and exclusions

The questionnaire was filled out by 92 children (44 in the affected group and 48 in the control group). Eight patients refused to participate in the study. All patients of the affected group had not been previously treated for their voiding problems. No child in the control group had voiding or defecation problems at the time they completed the questionnaire.

The age at presentation was similar in both groups ($p=0.86$) (Table 1). Boys accounted for approximately 60% of cases for each group ($p=0.5$).

Table 1. Patient's demographics and item scores

Parameter	Diseased group n=44	Control group n=48	p
Age at presentation mean (range)	117.8 months (78–167.9)	117 months (72.4–165.6)	0.86
Gender			
Male n (%)	26 (59.1)	29 (60.4)	0.5
Female n (%)	18 (40.9)	19 (39.6)	
BBDSS items			
Bladder issue			
Item 1 mean	1.34	0.04	<0.001
Item 2 mean	1.3	0.1	<0.001
Item 3 mean	1.43	0.33	<0.001
Item 4 mean	0.68	0.02	<0.001
Item 5 mean	0.23	0.02	0.009
Item 6 mean	0.73	0.04	<0.001
Item 7 mean	1	0.3	<0.001
Total mean (range)	6.7 (3–11)	0.88 (0–3)	<0.001
Bowel issue			
Item 1 mean	0.61	0.17	<0.001
Item 2 mean	0.3	0.1	0.05
Item 3 mean	0.5	0.02	<0.001
Item 4 mean	0.39	0.06	0.001
Item 5 mean	0.23	0.02	0.005
Total mean (range)	1.94 (0–6)	0.37 (0–4)	<0.001
Total BBDSS score mean (range)	8.7 (3–14)	1.19 (0–5)	<0.001

BBDSS: Bladder Bowel Dysfunction Scoring System.

BBDSS evaluation

Of all the children assessed, two (2.1%) found the questionnaire language to be difficult and four (4.3%) almost understood the questionnaire. The remaining children (93.6%) had no difficulties answering the questionnaire. The mean total score for the affected group was 8.7 (3–14), while it was 1.19 (0–5) for the control group ($p<0.001$) (Table 1). For the voiding-specific section of the questionnaire, the mean score was 6.7 (3–11) for the affected group and 0.88 (0–3) for the control group ($p<0.001$). For the defecation-specific section, the mean score was 1.94 (0–6) for the affected group and 0.37 (0–4) for the control group ($p<0.001$). The scores for all individual questions of the BBDSS questionnaire were significantly different between both study groups.

There was a strong correlation between the total BBDSS score and both groups using the Pearson correlation coefficient ($r=0.88$, $p<0.001$). The voiding section was strongly correlated to both groups ($r=0.89$, $p<0.001$), while the defecation section was moderately correlated ($r=0.53$, $p<0.001$).

Discriminant function analysis indicated that the BBDSS questionnaire had 90% accuracy in classifying all included patients correctly.

Using the ROC curve, the BBDSS was found to be an excellent tool in differentiating normal and affected patients (area under the curve [AUC]=0.98, 99% confidence interval [CI] 0.97–1, $p<0.001$). When the total BBDSS score was ≥6, the positive predictive value was 1, while the negative predictive value 0.89 (Figure 2).

Within the affected group, 24 patients were diagnosed with BBD, while OAB was the working diagnosis for 20 patients. No item on the voiding section of the BBDSS was significantly different between the OAB and BBD subgroups (Table 2). With respect to the defecation section of the questionnaire, all questions differed significantly between OAB and BBD children except item 4 (painful defecation). The mean defecation score was 0.65 (0–2) for OAB patients, while patients with BBD has a mean score of 3.04 (1–6) (p=0.002). We evaluated the defecation section of the BBDSS using the ROC curve. The ROC curve (Figure 3) showed that the defecation part of the BBDSS was a good test in differentiating OAB from BBD patients (AUC=0.89, 99% CI 0.79–0.99, p<0.001). No patient with OAB had a bowel score >3. The cutoff value of 3 had a positive predictive value of 100% (CI 79.9–100%) and negative predictive value of 70.8% (CI 48.8–86.6%) for BBD diagnosis.

The internal consistency of the BBDSS questionnaire was evaluated using Cronbach's alpha. The BBDSS was found to have a moderate internal consistency of 0.51 when comparing the total score to the diagnosis. The internal consistency of the bowel section was 0.73 (good internal consistency), while the internal consistency for the voiding section was 0.57 (moderate internal consistency).

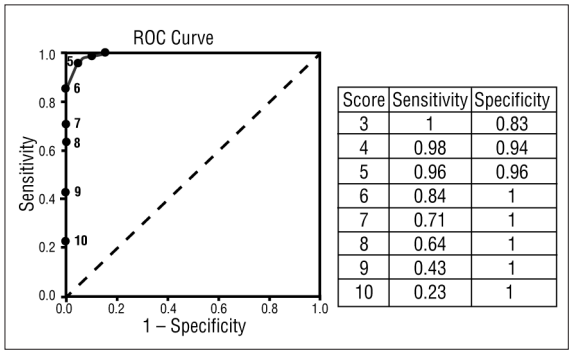
Table 2. BBDSS item scores in relation to the primary diagnosis of the diseased group

Parameter	Overactive bladder n=20	Bladder bowel dysfunction n=24	p
Bladder issue			
Item 1 mean	1.2	1.5	0.21
Item 2 mean	1.35	1.25	0.51
Item 3 mean	1.25	1.6	0.17
Item 4 mean	0.7	0.67	0.78
Item 5 mean	0.3	0.17	0.67
Item 6 mean	0.75	0.71	0.91
Item 7 mean	1.1	0.91	0.42
Total mean (range)	6.65 (3–10)	6.75 (3–11)	1
Bowel issue			
Item 1 mean	0.35	0.83	0.03
Item 2 mean	0	0.54	0.001
Item 3 mean	0	0.92	<0.001
Item 4 mean	0.25	0.5	0.28
Item 5 mean	0.05	0.38	0.02
Total mean (range)	0.65 (0–2)	3.04 (1–6)	<0.001
Total BBDSS score mean (range)	7.35 (3–10)	9.88 (5–14)	0.002

BBDSS: Bladder Bowel Dysfunction Scoring System.

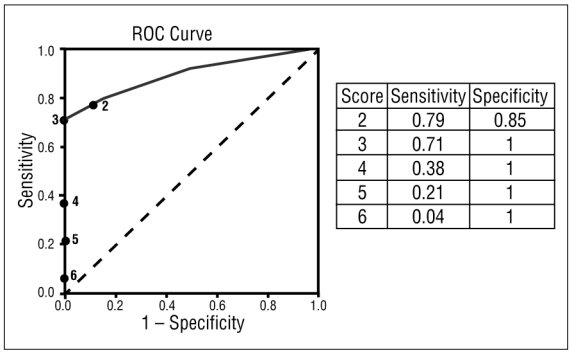
DISCUSSION

Despite being a common condition in the pediatric population, BBD is considered primarily a clinical diagnosis without a universal agreement on its diagnostic criteria. Healthcare professionals worldwide use different bladder and bowel questionnaires depending on their preferences. As a result, there is a strong need for a standardized questionnaire that can be used universally for the diagnosis and quantitative measurement of the severity and treatment response of BBD. Currently, there are only a few questionnaires available that concomitantly evaluate bladder and bowel symptoms, and their uses in clinical settings have limitations.[1,4,8] Other questionnaires address either bladder or bowel dysfunction in isolation.[2,3,9-14] Recently, new questionnaires were developed to better evaluate bowel function.[15,16]



Score	Sensitivity	Specificity
3	1	0.83
4	0.98	0.94
5	0.96	0.96
6	0.84	1
7	0.71	1
8	0.64	1
9	0.43	1
10	0.23	1

Figure 2. Evaluation of the Bladder Bowel Dysfunction Scoring System (BBDSS) in differentiating normal and diseased patients. Area under the curve=0.98 (confidence interval 0.97–1, p<0.001).



Score	Sensitivity	Specificity
2	0.79	0.85
3	0.71	1
4	0.38	1
5	0.21	1
6	0.04	1

Figure 3. Evaluation of the bowel questions of Bladder Bowel Dysfunction Scoring System (BBDSS) in differentiating overactive bladder (OAB) and bladder bowel dysfunction (BBD). Area under the curve=0.89 (confidence interval 0.79–0.99, p<0.001).

The Rome IV criteria were introduced in 2016 to replace the Rome III criteria for the diagnosis of functional gastrointestinal problems.[5] Rome IV criteria were used to evaluate the prevalence of functional constipation in the pediatric population.[17] These criteria demonstrated a good ability to diagnose functional constipation in a study group of 959 children. The definition of hard stool was not adequately interpreted by the children and parents in self-administrated questionnaires. The conjugation of the Bristol stool form scale, a pictorial diagnostic tool used to evaluate stool based on shape and consistency, could be expected to circumvent this limitation and improve the assessment of bowel function.

The quality of the urinary stream, despite its importance, has never been a focus of assessment in any questionnaire evaluating children with voiding dysfunction to date. In our opinion, it would be difficult to create a simplified and clear question in a self-administrated questionnaire without it being a detailed and likely cumbersome description of normal and abnormal urinary streams. Our idea was not new and had been previously presented in the visual prostate symptom score questionnaire.[18] We, therefore, chose to use simple illustrations to help distinguish normal and abnormal urinary streams.

After revision of the literature, various BBD questionnaires are illustrated in Supplementary Table 1 (available at *cuaj.ca*). The first proposed instrument for voiding assessment in children is the Dysfunctional Voiding Score System (DVSS), which is still commonly used worldwide today.[19] This questionnaire, however, was extracted from the International Prostate Symptom Score, which could be inappropriate in children. In addition, this instrument lacks reliability assessment, an important parameter in questionnaire standardization. Furthermore, important lines of questioning suggestive of bowel dysfunction, such as fecal soiling, are lacking in the questionnaire. Inadequate information concerning a child's bowel habits could clearly lead to inappropriate management. Lastly, there is no available assessment of responsiveness for the DVSS.

In 2009, Afshar et al constructed the "Vancouver scale" to evaluate BBD, with excellent test-retest reliability and validity.[4] This questionnaire was composed of 10 questions for bladder symptoms and three for bowel symptoms. The questionnaire was further validated in the pediatric urology clinic.[20] The questionnaire's internal consistency is poor (Chronbach α=0.45). Furthermore, this questionnaire has not been assessed for responsiveness. Therefore, similar to the DVSS, the Vancouver scale has not yet been validated to measure the treatment response of BBD.

Van Engelenburg-van Lonkhuyzen proposed a parent-reported 18-item Childhood Bladder and Bowel Dysfunction Questionnaire (CBBDQ) that had good internal consistency for both bladder (Chronbach α=0.74) and bowel (Chronbach α=0.71) subscales.[7] The questionnaire is currently in its early stage of evaluation. Important measurement properties, such as test-retest reliability, responsiveness, and interpretability, are to be evaluated in the subsequent phase.

Our questionnaire was developed with the intent to provide parents, children, and clinicians with a universal instrument to guide in the accurate diagnosis and evaluation of the evolution of BBD in a patient. Our main goal was to develop a reliable and valid instrument that could be completed with ease by children and their parents. We developed the first questionnaire with illustrations to help answer questions on both urinary stream and stool quality. Stool quality was illustrated according to the Bristol stool chart, which is now considered a valuable tool in the diagnosis and evaluation of treatment response for bowel dysfunction.[6,19,21] The illustrations also played an important role in engaging the children to fill out the questionnaire. Overall, most participants in the study considered the questionnaire easy to complete, with language appropriate for children aged 6–14. The questionnaire was completed within 3–5 minutes prior to their visit to the clinic.

Our analysis demonstrated the strong reliability and validity of the questionnaire in discriminating affected patients from those of the control group. All individual questions scored significantly different between the affected and control groups. Additionally, in terms of the evaluation of BBD, both voiding scores and bowel scores were significantly different between the affected and control groups. Furthermore, there was a strong correlation between the total BBDSS score and both groups. Finally, the ROC curve for the BBDDS demonstrated excellent differentiating power between affected and normal groups (AUC=0.98, 99% CI 0.97–1, $p<0.001$). With the cutoff of 6 in total score, we were able to differentiate the affected group from the control group with a positive predictive value of 1 and a negative predictive value of 0.89.

While the symptoms of OAB and BBD can overlap with respect to voiding symptoms, their treatments differ significantly. Urotherapy (education, adequate hydration, timed voiding, and pelvic floor muscle awareness) and constipation management remain the primary management strategies for BBD, while pharmacotherapy (anticholinergics) is the mainstay treatment for OAB.[19] Our questionnaire is the first instrument to be able to

significantly differentiate OAB from BBD patients. At the cutoff bowel score of 3, we were able to significantly differentiate OAB patients from BBD patients with a positive predictive value of 1.00 and a negative predictive value of 0.78. Overall, the questionnaire provided a strong discriminatory measure between OAB and BBD for the first time. Accurate diagnosis of these conditions is clearly an important prerequisite in guiding appropriate management strategy.

Limitations

The study design is not without limitations. The sample size was relatively small; however, the study is pilot and comparable to that of other studies.[2,4] A further study is warranted to evaluate more patients to confirm the usefulness of the BBDSS in the assessment and management of BBD.

The questionnaire was also only evaluated in a single institution and, therefore, would certainly benefit from evaluation at different institutions.

Furthermore, the evaluation of treatment response should be tested in further studies. The idea of conducting an OAB/BBD questionnaire is not new and recent questionnaires provided more questions about constipation depending on ROME criteria;[15,16] however, in our questionnaire, we tried to provide a tool that can help distinguish between OAB and constipation. As the questionnaire was administered only in English in our evaluation, translation to other languages will need to be carried out in future assessments.

CONCLUSIONS

This pilot study demonstrated that the BBDSS is a reliable, valid, and accurate instrument in the diagnosis of voiding dysfunction. The questionnaire is easily administrated by parents or children. Moreover, the defecation section of the questionnaire can differentiate between OAB and BBD with 100% positive predictive value.

COMPETING INTERESTS: The authors do not report any competing personal or financial interests related to this work.

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This paper has been peer-reviewed.

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