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Bladder and Bowel Dysfunction in Korean Children with Down Syndrome and Parental Quality of Life

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ABSTRACT

Purpose: Children with Down syndrome (DS) are more likely to experience bladder bowel dysfunction (BBD) than typically developing children, which could in turn have a serious effect on children with DS and on their parents and other family members. This study aimed to explore the prevalence of BBD in Korean children with DS and its effect on parental quality of life (QOL).
Design and methods: To assess BBD and parental QOL, we used self-administered questionnaires (Dysfunctional Voiding Symptom Score [DVSS], Rome IV criteria, and World Health Organization Quality of Life scale [WHOQOL-BREF]) for parents of children with DS. We collected data from 86 parents between September and October 2017 through an online community website.
Results: DVSS was elevated in 26.7% of the children with DS. Specifically, 14% had daytime urinary incontinence, and 33.7% had functional constipation. Moreover, 18.6% of children had BBD according to the DVSS and Rome IV criteria. The sensitivity and specificity of DVSS to functional constipation was 55.17% and 87.72%, respectively. The BBD score and total parental QOL score were statistically correlated ($r = 0.291$, $p = 0.007$).
Conclusions: Although children with DS are a high-risk group for BBD, their BBD symptoms are often overlooked because of their intellectual disability. Consequently, this could negatively affect children's and family's health and QOL in the long term.
Practice implications: Health-care providers should reconsider a routine check-up of BBD in children with DS. If a child has BBD, health-care providers should consult a urologist to determine the appropriate diagnosis and intervention.

Introduction

Bladder bowel dysfunction (BBD) is a spectrum of lower urinary tract symptoms associated with bowel complaints (Austin et al., 2016). Because of the close relationship between the bladder and bowel, which is attributable to their common innervation and pelvic floor muscle, children often present with BBD (Aguiar & Franco, 2018). Therefore, BBD is an umbrella term that can be subcategorized into lower urinary tract (LUT) dysfunction and bowel dysfunction (Austin et al., 2016). The true incidence of BBD is unknown; however, it is estimated that the overall prevalence rates of daytime urinary incontinence, which is one of the severe symptoms of BBD, are 10.5–11% at 4.5 years and 2.2–3.2% at 9.5 years (Heron, Joinson, Croudace, & von Gontard, 2008), and functional constipation prevalence is 14.1% in children older than 4 years of age (Robin et al., 2018). BBD is not confined to childhood. The link between childhood LUT symptoms and persistent adult problems with overactive bladder has been discussed in previous studies (Fitzgerald et al., 2006; Franco, 2016; Minassian, Lovatsis, Pascali, Alarab, & Drutz, 2006; Stone, Rozzelle, & Greenfield, 2010). Additionally, one study showed that approximately 50% of children continue to experience constipation 10 years after the initial complaint and that 25% of children had involuntary soiling (Michaud, Lamblin, Mairesse, Turck, & Gottrand, 2009). Moreover, 25% of children exhibit BBD symptoms until adulthood (Rajindrajith, Devanarayana, & Benninga, 2013).

Down syndrome (DS) is the most common chromosomal disorder associated with intellectual disability (Perluigi & Butterfield, 2012).

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Children with DS also have multiple malformations or medical conditions associated with their cardiac, ophthalmologic, hearing, thyroid, and gastrointestinal systems (Bull & Committee on G, 2011). However, BBD is not considered to be a common problem, and BBD in children with DS is often unrecognized or underdiagnosed because health-care providers and parents often assume bladder and bowel symptoms are part of the DS phenotype (de Carvalho Mrad et al., 2014), thereby they receive less attention (Kitamura et al., 2014). As people with DS live longer than expected, their families frequently report that they have LUT dysfunction, such as decreased voiding frequency and urinary incontinence (Kitamura et al., 2014). Consequently, several studies have investigated BBD in DS in the early 21st century (de Carvalho Mrad et al., 2014; Hicks, Carson, & Malone, 2007; Kitamura et al., 2014; Mrad, Figueiredo, Bessa, & Bastos Netto, 2018; Niemczyk et al., 2017; Powers et al., 2015). Urinary incontinence was reported in 46–68% of children with DS (Hicks et al., 2007; Niemczyk et al., 2017; Powers et al., 2015), and functional constipation was observed in 50% of children with DS (de Carvalho Mrad et al., 2014). These results indicate that children with DS are more likely to experience BBD than typically developing children.

Recent reports support the relationship between BBD and urinary tract infection, which could predispose a child to recurrent infection and thus potentially cause renal scarring (Keren et al., 2015; Shaikh et al., 2016). Moreover, BBD negatively affects family, school performance, and social relationships with typically developing children (Veloso, Mello, Ribeiro Neto, Barbosa, & Silva, 2016).

Furthermore, BBD in children with DS could also negatively affect parental quality of life (QOL). Parents of children with BBD experience significantly higher stress level, frustration, anger, depression, reduced parental QOL, and family conflict (Cushing et al., 2016; De Bruyne et al., 2009; Klages et al., 2017; Michaud et al., 2009; Thibodeau, Metcalfe, Koop, & Moore, 2013). Parents of children with DS have higher stress than parents of typically developing children because of high demands for care, economic problems, and social attitudes (Crnic, Arbona, Baker, & Blacher, 2009). Thus, parents of children with DS and BBD have significantly increased stress levels because of several reasons, which may also include increased laundry demands, financial costs, restrictions in going out, and stigma related to BBD (Collis, Kennedy-Behr, & Kearney, 2019). However, no study that focused on the relationships between BBD and parental QOL in children with DS has been conducted.

Hence, the purpose of this study was to explore the prevalence of BBD including LUT dysfunction and bowel dysfunction in Korean children with DS, and to explore its relationship with parental QOL.

Design and methods

Design and study subjects

This was a cross-sectional descriptive study involving 86 parents of toilet-trained children (those with voluntary control over their lower urinary function) with DS aged 4 to 18 years; the parents who are able to access to the Internet, read and write Korean, and agreed to participate in this study were included. To determine the appropriate sample size for this study, a power analysis was performed using G*power 3.1.9.2 (University of Düsseldorf, Düsseldorf, German). Overall, 82–90 participants were needed for a correlation analysis with a 0.05 α-level, 80% power, 0.3 median effect size, and 10% dropout rate (Cohen, 1992); therefore, our sample size of 86 participants was sufficient.

This study was approved by the (*Author blinded*) institutional ethics committee. The survey was posted on the online community website. This online community of parents of children with DS is the largest in South Korea with 3800 members and about 200 visitors per day and serves as a peer support group. Potential participants were provided with a link to the project information sheet where interested participants could access the online survey and where details of the study could be found. Informed consent was obtained online. The link was open from September 15 to October 24, 2017. The participants completed the questionnaire in approximately 10 min. We offered a mobile coffee coupon (valued at approximately 4 USD) to the participants as a token of gratitude for completing the survey.

Instruments

Dysfunctional Voiding Symptom Score (DVSS)

Children's BBD symptoms were determined using the well-established DVSS questionnaire (Farhat et al., 2000), which is helpful in obtaining objective evidence to support the diagnosis of BBD (Aguiar & Franco, 2018). It has been tested and validated across cultures and has undergone tests and re-tests for reliability (Farhat, McLorie, O'Reilly, Khoury, & Bagli, 2001; Lee, Farhat, & Park, 2014). The questionnaire consists of 9 items (2 items related to defecation, 7 items related to voiding) measuring the frequency of dysfunctional voiding in 1 month and 1 item measuring the stress recently experienced by children. A 4-point Likert scale was employed (0: almost none, 1: <50%, 2: ≥50%, 3: almost always, NA: not available), and the total number of points was 30 (the higher the score, the higher the BBD). In the study of Farhat et al. (2000), the cut-off DVSS was >9 points for boys and >6 points for girls. We used the validated Korean version of the DVSS (Lee et al., 2014), whose Cronbach's α was 0.97 and 0.704 in Lee's and Farhat's study (Farhat et al., 2000; Lee et al., 2014), respectively.

Rome IV criteria

To diagnose functional gastrointestinal disorders in children (≥4 years old), the Rome IV criteria were developed in 2016 (Hyams et al., 2016). In this study, we used the criteria on functional constipation. The presence of ≥2 of the 6 criteria (defecation ≤2 per week, at least 1 episode of fecal incontinence per week, history of retentive posturing or excessive volitional stool retention, history of painful or hard bowel movements, presence of a large fecal mass in the rectum, history of large diameter stool that could obstruct the toilet) for the past 1 month was considered functional constipation (Koppen, Nurko, Saps, Di Lorenzo, & Benninga, 2017).

World Health Organization QOL scale (WHOQOL-BREF)

Parents' QOL was assessed with the abbreviated version of the WHOQOL-BREF (in Korean) (Min et al., 2002), which has been cross-culturally adapted. The WHOQOL-BREF included 24 items across four domains (Cronbach's alpha for the total score was 0.963): physical health (7 items), psychological health (6 items), social relationships (3 items), and environment (8 items) and another 2 general items: self-evaluation of QOL and satisfaction with health. Each item, relating to the situations 2 weeks prior to the survey, is scored using a 5-point Likert rating scale (1 = least favorable condition to 5 = most favorable condition). The results were given as gross scores, which were subsequently converted into a score between 0 and 100; a higher score represents better QOL. Cronbach's α was 0.898 in the study by Min et al. (2002) and 0.952 in this study.

Demographic characteristics

There were 15 items concerning demographic characteristics of the children with DS and their parents. Parent characteristics included gender, age, marital status, education, occupation, and economic status. Child characteristics included gender, age, birth order, height, weight, type of enrolled education, and presence of other health problems. Health problems included but were not limited to congenital heart disease, thyroid disease, and hearing, vision, gastrointestinal, dental, musculoskeletal, nervous system, skin, and sleep problems (Bull & Committee on Genetics, 2011).

Statistical analysis

Statistical analyses were performed using IBM SPSS Statistics 23.0 (IBM, South Korea). Descriptive statistics, chi-square test, Fisher's exact test, *t*-test, and Pearson's correlation coefficient were used. Results were considered statistically significant at $p < 0.05$.

Results

The age of children with DS ranged from 5 to 17 years (8.1 ± 3.3 years; male 45.3%). Based on parental reports, approximately 62 (72.9%) children with DS had 1 to 3 health problems and 58.1% of children were enrolled in mainstreaming education. A total of 86 parents of children with DS participated in this study (mean age 41.1 ± 5.1 years, mothers 87.2%, graduated from college or graduate school 81.2%) (Table 1).

Table 2 demonstrates the prevalence rate of BBD symptoms and their differences according to presence of BBD, sex, and age group. Twenty-three (26.7%) children with DS had BBD based on the DVSS, and 14% of participants had daytime urinary incontinence. The mean DVSS was 11.9 ± 4.8 in children with DS with BBD and 3.6 ± 2.2 in children without BBD ($p < 0.001$). Specifically, daytime incontinence, frequency of evacuation, push bowel movements, decreased frequency of urination, holding maneuvers, urinary urgency, and straining voiding were significantly higher in children with DS with BBD than those without BBD. No difference between the BBD symptoms was noted with respect to the sex; however, daytime incontinence was statistically more prevalent ($p = 0.002$) in the 5–12-year age group than in the 13–16-year age group. However, no significant differences in BBD according to the general characteristics of children with DS (sex, age, birth order, body mass index, type of enrolled education, and number of health problems) were found (Table 3).

Table 1
Characteristics of respondents and children with Down syndrome ($N = 86$).

Variable	n (%)	M ± SD (range)
Children with Down syndrome		
Sex		
Boys	39(45.3)	
Girls	47(54.7)	
Age		8.1 ± 3.3 (5–17)
5 to 6 years	38(44.2)	
7 to 12 years	37(43.0)	
13 to 17 years	11(12.8)	
Birth order		
first	47(56.6)	
second	29(35.0)	
third	7(8.4)	
Body mass index (kg/m^2)[a]		17.7 ± 3.0 (13.2–27.2)
Under weight	6(7.0)	
Normal	57(66.3)	
Overweight	8(9.3)	
Obesity	15(17.4)	
Type of enrolled education		
General education	15(17.4)	
Mainstreaming education	50(58.1)	
Special education	21(24.5)	
Number of health problems		
None	16(18.8)	
1	26(30.6)	
2	24(28.2)	
3	12(14.1)	
>4	7(8.3)	
Parents		
Gender		
Father	11(12.8)	
Mother	75(87.2)	
Age(year)		41.1 ± 5.1 (31–54)
Married status		
Married	81(95.3)	
Single	4(4.7)	
Highest level of education		
Middle school	1(1.2)	
High school	15(17.6)	
Graduate school	59(69.4)	
Postgraduate school	10(11.8)	
Occupation		
Yes	38(44.7)	
No	47(55.3)	
Level of economic status		
High-Middle	14(16.3)	
Middle	47(54.7)	
Low-middle	18(20.9)	
Low	7(8.1)	

[a] Body Mass Index (BMI): Korean Centers for Disease Control and prevention & Korean Journal of Pediatrics (2017). Standard growth chart.

Cross-analysis of LUT dysfunction (based on a sex-specific cut-off score of the DVSS) and bowel dysfunction (≥2 of 6 of the Rome IV criteria were met) is shown in Table 4. LUT dysfunction was present in 26.7% of the children with DS, and 16 (18.6%) participants had BBD according to the DVSS and Rome IV criteria. The sensitivity and specificity of DVSS to functional constipation was 55.17% (95% confidence interval (CI) 35.69–73.55%) and 87.72% (95% CI 76.32–94.92%), respectively. There is a statistically significant association between the patient with >2 Rome criteria and DVSS that are considered elevated ($p < 0.001$). The odds of having a positive DVSS with ≥2 Rome criteria was 2.3 while the odds of having a positive DVSS with <2 Rome criteria was 0.26 leading to an odds ratio of 8.8.

Although the parents of children with DS with BBD based on DVSS had lower scores in all subcategories and lower QOL than those of children without BBD, no significant differences were noted. However, a significant negative correlation was observed between the BBD scores based on DVSS of children with DS and the physical ($r = -0.330$, $p = 0.002$), psychological ($r = -0.214$, $p = 0.047$), and environmental ($r = -0.246$, $p = 0.022$) domains, with the sole exception noted in the social relationship domain ($r = -0.210$, $p = 0.053$). Moreover, the total DVSS and specific overactive bladder symptoms were statistically correlated with the total parental QOL score ($r = -0.291$, $p = 0.007$ and $r = -0.422$, $p < 0.001$, respectively) (Table 5).

Discussion

We found that the DVSS was elevated in 26.7% of the children with DS. Moreover, 18.6% of the children had BBD, which means they had a positive DVSS and Rome IV criteria. Studies investigating BBD in individuals with DS are few (de Carvalho Mrad et al., 2014; Hicks et al., 2007; Kitamura et al., 2014; Mrad et al., 2018; Niemczyk et al., 2017; Powers et al., 2015), and to our knowledge, this is the second study that used the DVSS in children with DS. The prevalence of LUT dysfunction (based on the sex-specific cut-off score of the DVSS) in children with DS in this study was 26.7%, which is consistent with the result of a previous Brazilian study (de Carvalho Mrad et al., 2014) that included children with DS (27.3%). However, the prevalence of daytime incontinence was 14% (daytime incontinence according to DVSS = 1–3 points) in our study, which was significantly higher than that in previous studies: approximately 1.6% (Iran), 4% (Sweden), 6% (United Kingdom) for typically developing children (Safarinejad, 2007; Soderstrom, Hoelcke, Alenius, Soderling, & Hjern, 2004; Swithinbank, Heron, von Gontard, & Abrams, 2010). Conversely, the prevalence of daytime urinary incontinence among children with DS was much lower (14.0%) in this study than that in previous studies (46–68%) despite the similar age group of the enrolled children with DS (Hicks et al., 2007; Kitamura et al., 2014; Niemczyk et al., 2017; Powers et al., 2015). This discrepancy could be attributable to the different ways of defining patients with BBD based on the DVSS as these studies did not utilize the DVSS as did the present study and Brazilian study.

Table 2
Prevalence of bladder and bowel dysfunction (BBD) symptoms and differences according to the presence of BBD, sex, and age.

Variable	Presence of symptoms	Item score[a] (N = 86)	BBD			Sex			Age		
			With BBD (*n* = 23)	Without BBD (*n* = 63)	*p*-Value	Boy (*n* = 39)	Girl (*n* = 47)	*p*-Value	5–12 years (*n* = 75)	13–16 years (*n* = 11)	*p*-Value
1. Daytime incontinence	14.0%	0.21 ± 0.60	0.57 ± 0.99	0.08 ± 0.27	0.030	0.33 ± 0.77	0.11 ± 0.38	0.099	0.24 ± 0.63	0.00 ± 0.00	0.002
2. Volume of urine leakage	8.1%	0.13 ± 0.50	0.39 ± 0.89	0.03 ± 0.18	0.067	0.23 ± 0.71	0.04 ± 0.20	0.115	0.15 ± 0.54	0.00 ± 0.00	0.371
3. Frequency of evacuation	52.3%	0.93 ± 1.09	1.78 ± 1.24	0.62 ± 0.85	0.001	1.10 ± 1.17	0.79 ± 1.02	0.185	0.97 ± 1.13	0.64 ± 0.81	0.343
4. Push bowel movements	58.1%	0.98 ± 1.03	1.78 ± 1.13	0.68 ± 0.82	<0.001	1.03 ± 1.11	0.94 ± 0.97	0.691	1.03 ± 1.04	0.64 ± 0.92	0.242
5. Decreased frequency of urination	29.1%	0.51 ± 0.93	1.30 ± 1.26	0.22 ± 0.55	0.001	0.59 ± 0.99	0.45 ± 0.88	0.481	0.55 ± 0.96	0.27 ± 0.65	0.365
6. Holding maneuvers	38.4%	0.67 ± 1.00	1.35 ± 1.15	0.43 ± 0.82	0.001	0.64 ± 1.01	0.70 ± 1.00	0.780	0.67 ± 0.96	0.73 ± 1.27	0.852
7. Urinary Urgency	45.3%	0.78 ± 1.01	1.52 ± 1.16	0.51 ± 0.80	0.001	0.87 ± 1.08	0.70 ± 0.95	0.442	0.80 ± 1.01	0.64 ± 1.03	0.619
8. Staining voiding	33.7%	0.56 ± 0.90	1.43 ± 1.16	0.24 ± 0.50	<0.001	0.74 ± 0.99	0.40 ± 0.80	0.089	0.57 ± 0.89	0.45 ± 1.04	0.686
9. Dysuria	8.1%	0.16 ± 0.59	0.48 ± 0.99	0.05 ± 0.28	0.052	0.18 ± 0.64	0.15 ± 0.55	0.813	0.12 ± 0.49	0.45 ± 1.04	0.315
10. Stressful events	29.1%	0.87 ± 1.37	1.30 ± 1.52	0.71 ± 1.29	0.107	1.08 ± 1.46	0.70 ± 1.28	0.214	0.88 ± 1.38	0.82 ± 1.40	0.890
Total Score (possible score range: 0–30)	26.7%	5.80 ± 4.83	11.91 ± 4.78	3.57 ± 2.23	<0.001	6.79 ± 5.95	4.98 ± 3.52	0.098	5.97 ± 4.88	4.64 ± 4.55	0.395

[a] 0: almost none, 1: <50%, 2: ≥50%, 3: almost always.

Although urinary incontinence in children with DS is linked to multifactorial reasons (caregiver attitude toward the child, cognitive function, neuropsychiatric disturbances, and so on) (Powers et al., 2015), it is clear that toilet training in children with DS is markedly delayed (Mrad et al., 2018; Powers et al., 2015). In a previous study from United States, the average age at reported toilet training completion was 5.5 years in children with DS and 2.2 years in typically developing children. Among children 5 years or older, 79% of the children with DS were toilet trained, compared to the 100% of those without DS (Powers et al., 2015). In our study, urinary incontinence symptoms of participants were improved in the 13–18-year age group in contrast to the 5–12-year age group. Although our study enrolled participants who had completed toilet training, voiding control may have still not been completely developed at the age of 5–12 years.

The most dominant BBD symptoms in DVSS were related to constipation (DVSS for question 4 = 1–3; 58.1%), and around 33.7% had functional constipation based on Rome IV criteria. In a study by de Carvalho Mrad et al. (2014), they found that question 4 ("Does your son/daughter need to push for his/her bowel movements to come out?") was the most sensitive question, and if the answer to this question was no, it indicated a low probability of BBD. Furthermore, functional constipation according to Rome IV criteria was detected in 95.65% and 32.78% of children with and without LUTS, respectively (de Carvalho Mrad et al., 2014). Their findings are in accordance with our results that children with BBD had significantly higher scores for questions 4 and 5 than those without BBD. However, the sensitivity and specificity of DVSS to functional constipation in our study differed from those of de Carvalho Mrad et al. (2014). When comparing DVSS to Rome IV, the results showed that the sensitivity was low (55.17%) and the specificity was high (87.72%). Thus, if a child is negative in DVSS, he or she may not have functional constipation, whereas if a child is positive in DVSS, he or she is likely to have functional constipation; bowel habit evaluation for BBD should be recommended instead of overlooking the positive result, especially in children with LUTS (Aguiar & Franco, 2018; Yang et al., 2018).

Table 3
Differences of bladder bowel dysfunction according to general characteristics of children with Down syndrome.

Variable	With BBD[b]	Without BBD[b]	(N = 86)
	n (%)		x^2 (*p*)
Sex			0.044
Boys	10 (11.6)	29 (33.7)	(0.833)
Girls	13 (15.1)	34 (39.6)	
Age (years)[a]			1.089
5 to 6	9 (10.5)	29 (33.7)	(0.589)
7 to 12	12 (13.9)	25 (29.1)	
13 to 17	2 (2.3)	9 (10.5)	
Birth order[a]			2.683
First	13 (15.1)	36 (41.8)	(0.271)
Second	6 (7.0)	23 (26.7)	
Third	4 (4.7)	4 (4.7)	
Body Mass Index[a]			2.648
Under weight	3 (3.5)	3 (3.5)	(0.478)
Normal	16 (18.6)	41 (47.7)	
Overweight	1 (1.2)	7 (8.1)	
Obesity	3 (3.5)	12 (13.9)	
Type of enrolled education[a]			0.721
General education	5 (5.8)	10 (11.6)	(0.750)
Mainstreaming education	12 (14.0)	38 (44.2)	
Special education	6 (7.0)	15 (17.4)	
Number of health problems[a]			2.019
None	2 (2.3)	14 (16.3)	(0.379)
1 to 3	19 (22.1)	44 (51.2)	
>4	2 (2.3)	5 (5.8)	

[a] Fisher's exact test.
[b] Cut off score of BBD was above 9 points for boys and 6 points for girls.

There are several factors associated with BBD, including age and gender (Franco, Austin, Bauer, Von Gontard, & Homsy, 2015). Studies have shown a higher BBD prevalence in girls than in boys (Kyrklund, Taskinen, Rintala, & Pakarinen, 2012; Swithinbank

Table 4
Cross-analysis of bladder and bowel dysfunction.

Variable		Bowel dysfunction[a]		Total	Odds	OR[b]	χ^{2c}	p
		No	Yes					
LUT[d] dysfunction[e]	No	50(58.1%)	13(15.1%)	63	2.3	8.8	18.05	<0.0001
	Yes	7(8.1%)	16(18.6%)	23	0.26			
Total		57	29	86				
Sensitivity				55.17% (95% CI: 35.69% to 73.55%)				
Specificity				87.72% (95% CI: 76.32% to 94.92%)				
Positive likelihood ratio				4.49 (95% CI: 2.09 to 9.68)				
Negative likelihood ratio				0.51 (95% CI: 0.34 to 0.77)				
Disease prevalence				33.72% (95% CI: 23.88% to 44.72%)				
Positive predictive value				69.57% (95% CI: 51.48% to 83.12%)				
Negative predictive value				79.37% (95% CI: 71.74% to 85.18%)				

Sensitivity: a/a + c, Specificity: d/b + d, Positive predictive value: a/a + b, Negative predictive value: d/c + d, Positive likelihood ratio: Sensitivity/100-Specificity, Negative likelihood ratio: 100-Sensitivity/Specificity, Prevalence: (a + d)/a + b + c + d.
[a] Rome criteria.
[b] OR: Odds ratio.
[c] Fisher's exact test.
[d] Lower urinary tract.
[e] DVSS score.

Table 5
Correlation of BBD types based on the DVSS and parents' QOL (N = 86).

	Variable	X_1	X_2	X_3	X_4	X_5	X_6	X_7	X_8	X_9
X_1	The total score of DVSS	1								
X_2	Overactive bladder (item number 1 & 7)	0.671(<0.001)	1							
X_3	Dysfunctional voiding (item number 8 & 9)	0.732(<0.001)	0.321(0.003)	1						
X_4	Constipation (item number 3 & 4)	0.654(<0.001)	0.164(0.132)	0.450(<0.001)	1					
X_5	Physical QOL of parents	−0.330(0.002)	−0.425(<0.001)	−0.137(0.209)	−0.026(0.814)	1				
X_6	Psychological QOL of parents	−0.214(0.047)	−0.370(<0.001)	0.003(0.978)	−0.004(0.973)	0.792(<0.001)	1			
X_7	Social relationships QOL of parents	−0.210(0.053)	−0.454(<0.001)	−0.014(0.897)	−0.038(0.731)	0.626(<0.001)	0.643(<0.001)	1		
X_8	Environmental QOL of parents	−0.246(0.022)	−0.333(0.002)	−0.072(0.510)	0.039(0.718)	0.735(<0.001)	0.711(<0.001)	0.541(<0.001)	1	
X_9	Total QOL of parent	−0.291(0.007)	−0.422(<0.001)	−0.072(0.509)	−0.023(0.835)	0.872(<0.001)	0.902(<0.001)	0.773(<0.001)	0.836(<0.001)	1

BBD: bladder and bowel dysfunction, DVSS: dysfunctional voiding symptom score, QOL: quality of life.

et al., 2010) and demonstrated that in typically developing children, the prevalence of daytime incontinence decreases with age, which is consistent with our results, as 2 children had BBD after 13 years of age in contrast to 21 children in the 5 to 12 years age group. A previous study of children with DS showed that all BBD symptoms based on DVSS were more frequent in males than in females (de Carvalho Mrad et al., 2014). Conversely, no significant difference according to the sex was found in our study. However, we need to be cautious while interpreting the results of both studies because the participants' median age was 16 years (range 9–22) in that study (de Carvalho Mrad et al., 2014), while the mean age in our study was 8 (5–17) years.

In this study, the higher the BBD score based on DVSS of children with DS, the lower quality of life of the parents. Especially, overactive bladder symptoms including urinary urgency and daytime incontinence had a statistically significant negative relationship with parental QOL. These results are consistent with the results of studies by typically developing children with urinary incontinence and their parents (Collis et al., 2019; Equit, Hill, Hubner, & von Gontard, 2014). Parents of children with BBD had lower scores in all subcategories and total QOL than parents of those without BBD, although no significant differences were noted. The total QOL score of parents who have children with BBD was 55.28 ± 17.10 points. In previous studies using the same assessment tool, parental QOL of typically developing children with functional incontinence was 66.8 ± 15.6 and 67.7 in a control group (Equit et al., 2014). Moreover, the QOL score of parents of children with diabetic mellitus was 66.2 points (Park et al., 2015) and that of parents of children with hearing loss was 55 points (Ramires, Branco-Barreiro, & Peluso, 2016), indicating that QOL of parents of children with DS was lower than that of parents of typically developing children and parents with other chronic diseases and disabilities. Collis et al. (2019) described the significance of the effect of BBD, specifically in relation to burden of care, stigma, and social isolation of parents and children. They also reported on parents' perceptions of lack of health-care providers' advice and support (Collis et al., 2019). In children with disabilities who present with other primary and more evident secondary diagnoses, it is important that health-care professionals assess for BBD symptoms and provide the appropriate information, diagnosis, and treatment.

To our knowledge, this is the first study focusing on BBD in children with DS and parental QOL. Considering that the life expectancy of individuals with DS is approximately 60 years (Perluigi & Butterfield, 2012), prevention, early detection, and proper treatment of BBD is vital to prevent future health problems and social costs. Furthermore, currently, the goal of managing disabilities in children is shifting toward improving the QOL of both the children and their family (Perluigi & Butterfield, 2012); hence, BBD should be carefully evaluated.

Limitations

The limitations of this study include its cross-sectional design, which limits the analysis of causal effect. Furthermore, we used only questionnaires without clinical examinations to assess BBD symptoms, which was a methodological limitation. Moreover, as the number of adolescents enrolled in this study was small, the results should be interpreted cautiously, and our results should only be applied to children with DS because adults with DS may have different BBD symptoms. Lastly, there are limitations related to the online questionnaire, which was completed by the parents. There are no data available to determine whether the sample is representative and which major selection effects are present. Additionally, Internet samples are likely to be more biased than samples drawn from the general population (Strickland, 2017).

Implications for practice

Although health supervision for children with DS has been recommended by the American Association of Pediatrics since 2011 (Bull and Committee on G, 2011), there is no recommendation of screening for routine bladder and bowel symptoms in children with DS. We believe that these results could make health-care providers reconsider a routine check-up of BBD using a validated screening instrument such as DVSS in toilet-trained children with DS. Additionally, if a child has a positive DVSS, health-care providers should further evaluate his/her bowel habits using Rome IV. Then, if a child has functional constipation, health-care providers can treat it first according to the International Children's Continence Society's guideline (Yang et al., 2018). If the DVSS is positive despite resolution of functional constipation and gas, health-care providers should consult a urologist to determine the appropriate diagnosis and intervention. Especially, clinical nurses should be mindful of the consequences of LUTS, which are depicted mainly by urinary infections, injuries of the upper urinary tract (de Carvalho Mrad et al., 2014), and poor QOL of children with DS and their parents, as these issues could be prevented by educating parents about the symptoms of BBD and the importance of early detection.

Conclusions

Although children with DS are a high-risk group for BBD, their BBD symptoms are often overlooked because of their intellectual disability. This could in turn negatively affect the health and QOL of the children and their family in the long run. Based on a detailed assessment, BBD could be treated effectively; thus, the negative effects on activities of daily living of those affected and of parents and caregivers could be reduced. Therefore, health-care providers should pay attention to BBD symptoms and their management in children with DS.

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CRediT authorship contribution statement

Eun Kyoung Choi: Conceptualization, Methodology, Data curation, Formal analysis, Investigation, Supervision, Funding acquisition, Writing - original draft, Writing - review & editing. **Yong Ju Lee:** Methodology, Writing - review & editing. **Hooyun Lee:** Project administration, Resources. **Eunyoung Jung:** Data curation, Investigation, Formal analysis, Methodology, Writing - original draft.

Declaration of competing interest

None.

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