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A 2-Step Integrative Education Program and mHealth for Self-Management in Korean Children with Spina Bifida: Feasibility Study

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ABSTRACT

Purpose: The aim of this study was to develop and test the feasibility of a 2-step self-management program, including onsite integrative education and a mobile health (mHealth) intervention, for children with spina bifida (SB).
Design and methods: This feasibility study used a quasi-experimental single group pre-and post-test design. The onsite integrative education and the mHealth program, "Glowing Stars™," were developed and then tested for content validity by a panel of experts. The feasibility and user satisfaction were evaluated using factors such as school adjustment, self-management knowledge, self-efficacy, self-management behavior, and quality of life measurement by children aged 10 to 12 years with SB and their parents, from March 2018 to April 2018.
Results: A total of five children with SB completed this intervention. All children perceived that this program was usable and feasible to maintain self-management behavior. A statistically significant difference was observed in the children's self-management behavior domain between the first and second post-test ($p = .043$).
Conclusion: This innovative 2-step self-management intervention program complements existing single interventions and confirms the possibility of mHealth technology as an intervention for children with SB.
Practice implications: In pediatric nursing, this innovative intervention could be adapted for children with chronic conditions, with a positive effect on self-management.

Introduction

Spina bifida (SB) is the most common congenital neural tube condition (Wiener et al., 2017). SB occurs because of incomplete closure of the neural tube, occurring at approximately 28 days of gestation (Phillips, Burton, & Evans, 2017). Due to the improved management of children with SB over the past five decades (Szymanski, Cain, Hardacker, & Misseri, 2017), nearly all children with SB now survive into adulthood (Wiener et al., 2017). However, inappropriate or inadequate self-management behaviors may reduce the benefits of treatment and positive health outcomes, and increase the risk of secondary health outcomes (Stinson, Wilson, Gill, Yamada, & Holt, 2009); therefore, well-established self-management behaviors must be encouraged prior to adulthood (Lindsay, Kingsnorth, Mcdougall, & Keating, 2014).

Children with spina bifida and self-management

Self-management is important for people with SB (Copp et al., 2015; Stiles-Shields et al., 2019). In our previous need assessment of the parents of children with SB, across all ages, parents reported the need for self-management programs for their children (Choi, Ji, Bae, & Jang, 2019). The development of self-management competence in people with SB of school age is critical. According to the Life Course Model for SB (Swanson, 2010), self-management programs should be initiated during the preschool years and then applied efficiently to school-aged children, enabling patients to be independent by adolescence. By school age children with SB are expected to have acquired bowel and bladder self-management behaviors. These behaviors include clean intermittent self-catheterization (CIC), assisting the parent(s) in completing the bowel program, cleaning up after a bowel or bladder accident, and recognizing the signs of bowel problems and urinary tract infections (Greenley, 2010). Spina bifida management and monitoring of health needs are shared responsibly during childhood, with the child steadily taking on more responsibility for the management of his or her health (Gall, Kingsnorth, & Healy, 2006; Swanson, 2010). Although most children with SB achieve basic self-management and independence, they

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often lag behind their typically-developing peers by two to five years in these behaviors (Davis, Shurtleff, Walker, Seidel, & Duguay, 2006). Donlau et al. (2011) found that only 48% of children aged 12–18 in their study could complete the multiple component self-catheterization behavior. Components included keeping track of when to perform the CIC, preparing equipment, undressing as needed, transferring as needed, washing hands, preparing and inserting the catheter, notifying suppliers of catheters when stocks were low and ordering supplies needed for the CIC (Donlau et al., 2011). Medical non-adherence appeared to peak during late childhood for many medical domains (e.g., bowel programs, diet, exercise, and skin checks) (Psihogios, Kolbuck, & Holmbeck, 2015).

Self-management intervention for children with spina bifida

Several camp-based intervention studies to improve self-management for youths with SB have been published in the past 10 years (Driscoll et al., 2019; Holbein et al., 2013; O'Mahar, Holmbeck, Jandasek, & Zukerman, 2010; Zimmerman et al., 2019). They have reported significant improvements in SB self-management, but also many limitations. Limitations included improvements not maintained beyond the 1-month post-intervention period (O'Mahar et al., 2010), and lack of control groups (Driscoll et al., 2019; Holbein et al., 2013; O'Mahar et al., 2010; Zimmerman et al., 2019). Recently, a mobile health (mHealth) system named iMHere was developed and evaluated among adults with SB (Dicianno et al., 2016; Fairman et al., 2013; Parmanto et al., 2013; Yu, Parmanto, Dicianno, & Pramana, 2015; Yu, Parmanto, Dicianno, Watzlaf, & Seelman, 2017). However, this mHealth program was designed for adults only and was only effective among frequent users of the system in the feasibility test (Dicianno et al., 2016). There is general agreement that mHealth interventions are more effective than usual care, but almost half (43%) of the randomized controlled trials showed negative or unclear results for mHealth interventions (Chen, Chai, Dong, Niu, & Zhang, 2018). Therefore, for children, tailored, interactive, age- and development-appropriate, and condition-specific applications are recommended (Nightingale et al., 2017). mHealth for children need to be designed to include clear goals and the use of mHealth should instill a sense of achievement in the child (Dicianno, Henderson, & Parmanto, 2017).

Conceptual framework

In the Individual and Family Self-Management Theory (IFSMT) (Ryan & Sawin, 2009), self-management can be explained as a complex and dynamic phenomenon, consisting of contextual, process, and outcome dimensions. It is important to identify the pertinent contextual and process-related factors in the lives of children with SB to promote their readiness for and competence in self-management behaviors (Ryan & Sawin, 2009). We adapted this theory because of the role of self -management intervention on the context (such as school adjustment) and process (such as acquisition of skills, knowledge, and self-efficacy) of self-management and its impact on outcomes (including self-management behavior and quality of life).

To the best of our knowledge, this is the first study on the development and feasibility testing of a 2-step self-management program for school-aged children with SB. We hypothesize that new, theory-based intervention programs could improve on the shortcomings of camp-based interventions, and the development of a mHealth program could facilitate the sustained use of customized self-management programs for children with SB.

Purpose

The purpose of this study was to develop and test the feasibility of a 2-step self-management program, including onsite integrative education and a mHealth program for children with SB.

Design and methods

Design

This feasibility study used a quasi-experimental single group pre- and post-test design.

Participants

This study was conducted at the largest SB clinic in Seoul, South Korea using a convenience sampling technique. Participants were children with SB and their parents. For children with SB, the eligibility criteria included the following: (1) elementary school children (aged 7 to 12 years) with SB, (2) ability to use smartphones or use smartphone with the help of their parents, (3) absence of cognitive impairments, and (4) voluntarily consent to participate in this study. The exclusion criteria for the children included: (1) unable to communicate and respond to the questionnaires, (2) having illness or disabilities other than SB. The parents were included based on whether their children met the inclusion or exclusion criteria, and whether they were able to communicate and respond to the questionnaires and voluntarily consent to participate in the study.

The choice of sample size was based on previous studies that found that 80% of feasibility problems can be conducted with only five participants (Turner, Lewis, & Nielsen, 2006) and evaluation of accessibility of mHealth for people with SB (Yu et al., 2015). A drop-out rate of 20% was considered for this interventional research for school-aged children (Tanner-Smith & Wilson, 2013). Six children with SB and their parents were initially enrolled for the study. One child and his mother dropped-out and 5 children with SB and their parents completed this program.

Ethical considerations

Before conducting this study, ethical approval was obtained from the Institutional Review Board of the Yonsei University Health System. Voluntary participation, anonymity, and confidentiality were addressed. All participants were informed that they had the right to decline participation at any point. Details of the study were provided and written informed consent was obtained at a seminar room of a Yonsei University.

Development of the 2-step self-management intervention

To develop the 2-step self-management program, the following components were developed sequentially: onsite integrative education program and mHealth program.

Onsite integrative education program

Robust, user-led, well-developed information and support interventions are needed to address school-aged children's identified needs and preferences, while conforming to parents' and health care providers' views on what is realistic and achievable (Nightingale et al., 2017). Based on a literature review, we adapted the guidelines for the care of people with SB by the Spina Bifida Association of American (2018), the IFSMT, the Life Course Model (Swanson, 2010), and findings of our previous study on the need assessment of parents of children with SB (Choi et al., 2019) Based on the IFSMT, the contents of the current intervention program were developed to diminish possible risk factors and strengthen protective factors experienced by children with SB within the school environment (context), as well as improve self-management knowledge regarding SB (process).

Two authors (Choi and Ji) who are pediatric nurse practitioners (with 7 and > 10 years of experience in caring for children with SB) developed the contents of the onsite integrative education program. These authors have experience in caring for children with SB as pediatric nurse

practitioners, and they had previously developed the transanal irrigation bowel management program (Choi, Shin, Im, Kim, & Han, 2013; Choi, Han, Shin, Ji, Chon & Im, 2015) and a SB camp for school-aged children in Korea.

The onsite integrative education program for children with SB was developed as a 4 hour and 30-minute half-day course in 6 sessions. Table 1 presents the content and process of the onsite integrative education program. The intervention involved multiple teaching methods (including a lecture and discussion), laboratory exercise (using a human anatomy model), as well as role play and group activities (using a board game that addressed aspects of family life, school life, friendship, and themselves). This intervention was directed toward school-aged children with SB (Fig. 1).

After developing the program, a content validity test examining all used instruments was conducted by an expert panel of eight to identify suitability. The experts consisted of three pediatric urologists, three child health nursing professors, and pediatric nurse practitioners, who had all been directly involved in or had expertise in caring for children with SB. In this study, the content validity index of all intervention session exceeded 0.8, which was considered acceptable (Polit & Beck, 2017). However, several expressions were modified as per the expert's opinions.

mHealth program: 'Glowing Stars™'

Based on the contents of the onsite integrative education program, we developed an android mobile application with the name "Glowing Stars™" by selecting health indicators to be measured in order to maintain self-management skills. The health indicators of Glowing Stars™ for self-management behaviors included self-monitoring of voiding, defecation, skin care, taking medication and mood status. Our research team selected a mobile application developer with experience in medical-related application development. The steps involved the design of a system diagram, data models and database, an algorithm, and screens and menus. This was carried out by our research team and the application developer. We highlighted the significant need for mHealth gamification with the aim to optimize the maintenance of patient self-management behaviors using star-charts and statistical graphs (Fig. 2). Furthermore, we uploaded the lecture presentation files of an onsite integrative education program in Glowing Stars™ for referencing whenever necessary, or to share the contents with their parents.

The Glowing Stars™ application was developed using the Ionic 4 Framework (powered by Cordova and Google's Angular), with node.js 8.11. Graphs and charts were rendered by eChart, which supports multi-dimensional data visualization and various types of device screen size. Data between clients and server was encrypted and stored on a server configured using Amazon Web Services.

The Glowing Stars™ application was evaluated for content validity by the same experts involved in the content validity process of the onsite integrative education program. The content validity index (CVI) exceeded 0.8 in each category, which was considered acceptable (Polit & Beck, 2017).

Measurements

Measures used in the study were categorized as context, process, or outcome variables based on the IFSMT. The children reported on the context variables (school adjustment) and the process variables (self-

Table 1
Contents of the onsite integrative education program.

Session (time)	Theme/sub-theme	IFSMT component	Contents	Method
I (30 min)	Integrative education program (IEP) 1. Introduction 2. Understand the IEP	 Process	 1. Exchange greeting and self-introduction 2. Introduce the program purpose and process	 Discussion
II (30 min)	Spina bifida and self-management of voiding 1. Identify about SB 2. Know about voiding 3. Method of self-management for voiding	 Process Process Process	 1. Learning about SB 2. Sharing about my SB 3. Difference of voiding function between normal and SB condition 4. Consequence of voiding dysfunction (CIC, UI) 5. Voiding condition if not well managed in SB 6. CIC procedure, management of UI, medication, importance of water intake	 Lecture (PPT, video, quiz) Discussion
III (20 min)	Self-management of defecation 1. Know about Defecation 2. Defecation disorder 3. Prevention of defecation disorder	 Process Process Process	 1. Difference of defecation between normal and SB condition 2. Consequence of defecation disorder (Constipation, FI) 3. Defecation disorder in SB 4. Regular defecation habit, active management of defecation (medication, enema, defecation diary)	 Lecture (PPT. video, quiz) Discussion
IV (20 min)	Self-management of skin 1. Deformity of feet 2. Importance of skin management 3. Method of self-management for skin	 Process Process Process	 1. Differences in the shape of normal and deformed feet 2. Need for orthosis with deformed feet 3. Importance of skin management in the use of orthosis and wheelchairs 4. Regular skin observation 5. Reporting of abnormal skin conditions to your parent and healthcare provider 6. Prevention of pressure sores with the use of a wheelchair	 Lecture (PPT, quiz) Discussion
V (60 min)	Practice for voiding & defecation 1. Practice of urinary catheterization 2. Practice of enema	 Context & Process Context & Process	 1. Knowing about urinary catheterization and practicing using body manikins 2. Knowing about enema and practicing using a rectum model (i.e. *trans*anal irrigation, anal plug)	 Laboratory exercise
VI (60 min)	Talk Talk Talk!!! 1. Sharing a concerning situation and proposing the solution (Children of SB-led) 2. Closing the program	 Context & Process	 1. General concerning situations of spina bifida conditions (i.e., at school, at home, with peers, myself) 2. Sharing my experience in the situation and proposing a solution 3. Celebration and summary of the program	 Role play & group activity (board game format)

Note: CIC = clean intermittent catheterization, FI = fecal incontinence, IFSMT = the Individual and Family Self-Management Theory, PPT = PowerPoint, UI = urinary incontinence.

Fig. 1. Onsite integrative education program (Left: Self-management for defecation; Middle: Lab-exercise of self-management for clean intermittent catheterization; Right: Lab-exercise of self-management for transanal irrigation and anal plug).

management knowledge and self-efficacy). Their parents reported on the outcome variables, such as self-management behavior and quality of life.

School adjustment

Children's school adjustment was measured using a 20-item tool, which was developed by Kim (2002). It included teacher relations, peer relations, class lessons, and school regulations and was measured on a 6-point Likert scale, with 1 point for "Strongly disagree" to 6 points for "Strongly agree." Higher scores indicated a better degree of adjustment to school life. Cronbach's alpha range was from 0.72 to 0.80 in Kim's (2002) study.

Self-management knowledge

To measure children's self-management knowledge, we used Yun and Kim's (2017) self-management knowledge scale. The scale comprised of 18 questions to measure knowledge of self-management skills and condition-related knowledge of children with SB (Yun & Kim, 2017). Responses were measured using a 3-point scale, with 0 point for "Not applicable," 1 point for "No," and 2 points for "Yes." Higher scores indicated a higher degree of self-management knowledge. In Yun and Kim's (2017) research, Cronbach's alpha was 0.75.

Self-efficacy

Self-efficacy was measured using the general self-efficacy scale described by Chen, Gully, and Eden (2001), which consists of 8 questions on a 5-point Likert scale, with 1 point for "Strongly disagree" to 5 points for "Strongly agree." Higher scores indicated a higher degree of self-efficacy. Cronbach's alpha for this tool was 0.85–0.90 at the time of the tool development (Chen et al., 2001).

Self-management behaviors

Self-management behaviors were measured using the Kennedy Krieger Independence Scale-Spina Bifida Version (KKIS-S), which was developed by Zabel et al. (Zabel, Jacobson, Tarazi, & Mahone, 2012) and revised by Yun and Kim (2017). It was used to assess the parent-reported measure of the medical and self-management aspects of adaptive functioning and self-management competencies specific to SB. The KKIS-S is an 18–item scale with scores ranging from 18 to 54. Response options included "Yes", "Maybe yes", "Probably not", or "Not

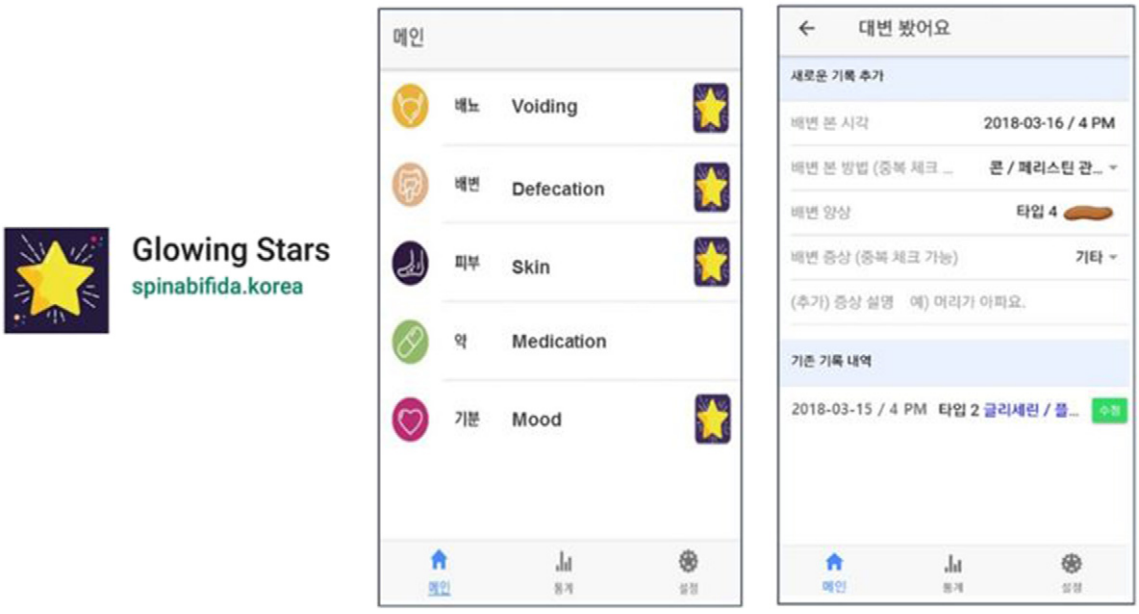

Fig. 2. Screen shot from the original (Left: the application listing as shown in the Google Play Store; Middle: The main screen, consisting of sections for voiding, defecation, skin, medication, and mood; Right: An example of the defecation section, including details such as time, method, type of Bristol stool scale, other symptoms and a memo regarding defecation in this time. The text below the heading at the bottom of the page represents a defecation history report).

applicable", with higher scores indicating greater independence of self-management. The Cronbach's alpha for this tool was 0.89 in Yun and Kim's (2017) study.

Health related quality of life (HRQOL)

Health-related quality of life (HRQOL) of children with SB was measured using the SB-HRQOL tool, developed by Parkin et al. (1997) for children and adolescents with SB and translated to the Korean language by Yun (2013). The Korean version of the HRQOL-SB consists of 44 items, with 8 questions rated on a 5-point Likert scale, with 1 point for "Only a little" to 5 points for "A lot". Higher scores indicated a higher health-related quality of life. The Cronbach's alpha for this tool was 0.93 at the time of tool development (Parkin et al., 1997) and 0.95 in Yun's study (2013).

Feasibility and user satisfaction evaluation

For the evaluation of the onsite integrated education program, we surveyed the children for their experience and satisfaction, through an open questionnaire about the onsite integrated education program given immediately after completion of the program.

For the evaluation of the mHealth program, participants used all the functions of our application daily for 4 weeks. Participants received the Uniform Resource Locator (URL) for the online survey (for feasibility evaluation) after completion of the mHealth program. The feasibility testing method for mHealth applications proposed by Jeon and Park (2018) in the development of apps for diabetes patients was used in this case. This method is composed of 18 items and measures 4 domains (quality of service, quality of system, satisfaction of users, and actual usage). It is measured on a 5-point Likert scale, with higher scores indicating higher quality in each domain.

Procedure and data collection

Data were collected between March and April 2018. Initially, participants (6 children with SB and their mothers) took three repetitive surveys over time using self-reported and parent-reported questionnaires delivered through online Google surveys. They fully completed the questionnaires on their own, so all participants were given complete privacy.

Pre-test was conducted 1 week before the onsite integrated education program. Only children with SB were enrolled in the onsite integrated education program, which was held in a seminar room and a simulation center at the Yonsei University College of Nursing. The two researchers who developed this program also conducted this program directly with the help of research assistants (Fig. 1). The first post-test was conducted immediately after the completion of the onsite integrated education program. Then, children with SB used the Glowing Stars™ mHealth program for 4 weeks at their homes on a daily basis. The mHealth Program allowed children to record and check self-management work daily, with help from their parents to monitor for appropriate use. The second post-test was conducted using an online survey, after 4 weeks of using the mHealth program. At the beginning of the program, we announced that they could complete this program successfully when they used the mHealth application for over 80% of 28 days (4 weeks). During mHealth intervention, a research assistant sent a text message to encourage daily recording if data had not been recorded over the previous 2 days. A research assistant also supported the children whenever they had questions about using the Glowing Stars™ app.

Following the onsite integrated education program, a token gift of a voucher worth 30,000 won (about 25 US dollars) was given to each participant. At the completion of the mHealth intervention and second post-test survey, we offered a mobile voucher worth 50,000 won (about 42 US dollars).

Data analysis

Data were analyzed using SPSS statistics software version 24.0 (IBM, Seoul, Korea). This included the descriptive statistics, a Wilcoxon's signed-ranks test, and repeated measures ANOVA analysis.

Results

Participant characteristics

Initially, six school-aged children who had regular follow-up care for SB management were recruited from the SB clinic. After the onsite integrated education program, one boy (10 years old) was unable to complete the mHealth intervention because he had only recorded a few days at the start of mHealth intervention and wanted to withdraw. Finally, five children completed the 2-step self-management program. All five children required CIC, and 40% wore pads due to urinary incontinence and/or fecal incontinence. Forty percent of the children needed additional methods to defecate regularly (such as laxatives or transanal irrigation) and 40% needed aids for mobility (Table 2).

Feasibility and user satisfaction

Feasibility evaluation was performed on the five children with SB and their parents, as shown in Tables 3 and 4. While commenting on the 2-step self-management program, children with SB expressed their satisfaction about knowing children with the same condition, and indicated that it motivated their self-management in the onsite integrated education program. The use of mHealth enabled them to check their status regarding self-management. Additionally, the parents requested some additional functions in the Glowing Stars™ app, such as a note function capable of recording memos relating to self-management procedures, and a feature allowing the recording of the menstrual period for girls (Table 3). In the cases of girls who had started menstruation, their parents felt their bladder and bowel symptoms were affected by menstrual period. Therefore, they wanted to record and compare those symptoms during their menstruation cycle. In the feasibility evaluation of mHealth by children with SB, more than a medium level of satisfaction was reported in all the domains; however, we needed to modify the aesthetic features of the app because this item had the lowest score (Table 4).

Modification of Glowing Stars™

We modified the Glowing Stars™ app based on the feasibility and user satisfaction evaluation. First, we added a note function for each domain. The note function allows the children to make a memo of specific symptoms as text in each subcategory. Secondly, we added menstrual period monitoring for girls in the voiding domain. Thirdly, we revised the starboard with a more attractive design to make it feel like a fairy tale book, in order to encourage users to check their records. Lastly, we added a function for sending their records to their email accounts as data files for printing based on comments made by parents.

Effects on self-management and quality of life

With respect to the five measures, there were no differences between the pre-test, and the first or second post-test, except that in the children's self-management behavior domain between the first and second post-tests ($p = .043$) (Table 5). In the children's self-management behavior, for three children, the scores dropped immediately after the onsite integrated education program but then rose after using mHealth for four weeks (Fig. 3).

Table 2
Characteristics of children with spina bifida (N = 5).

Children	Age	Gender	Voiding method	Defecation method	Mobility	Medication
Child 1	11	Female	Self-voiding, CIC with UI	Self-defecation	No aids	No
Child 2	11	Female	CIC with UI	Simple enema with FI	No aids	Yes
Child 3	12	Female	Self-voiding, CIC	Self-defecation	Wheel Chair	Yes
Child 4	12	Male	CIC	Transanal irrigation	Orthosis	Yes
Child 5	11	Male	CIC	Self-defecation	No aids	Yes

CIC = clean intermittent catheterization, FI = fecal incontinence, UI = urinary incontinence.

Discussion

To the best of our knowledge, this is the first study on the development and feasibility testing of a 2-step self-management program for school-aged children with SB. The 2-step self-management program that was developed in this study (including onsite integrated education and the mHealth program) had the following three characteristics: it was based on the innovation of a nursing intervention for children with chronic conditions, was an evidence-based intervention, and was a user-centered intervention.

Innovation of nursing interventions for children with spina bifida

Since the year 2000, various interventions have been developed for people with SB aimed at reducing the negative effects of SB and increasing the normalization of children with SB (Choi et al., 2019), especially through the development of camp interventions and transition clinics for adolescents and adults with SB (Driscoll et al., 2019; Fremion, Morrison-Jacobus, Castillo, Castillo, & Ostermaier, 2017; Holbein et al., 2013; O'Mahar et al., 2010; Zimmerman et al., 2019). Recently, mHealth interventions have been developed for adults with SB (Dicianno et al., 2016; Fairman et al., 2013; Parmanto et al., 2013; Yu et al., 2017). These various attempts to achieve self-management have several limitations. First, most interventions were for adolescents or adults with SB. Secondly, the camp intervention was a short-term, one-time intervention; therefore, its effects were difficult to maintain on a continuous self-management basis. Lastly, there has been no mHealth intervention for self-management in children with SB. Therefore, our 2-step self-management program improved on these shortcomings through developing an innovative nursing intervention for children with SB. In this program, children with SB could obtain self-management knowledge and share their experiences in order to enhance motivation and encourage continual self-management. These effects could also be obtained from a "one time" camp intervention; however, maintaining continuity is more important for self-management. Based on our study, a mHealth focus on maintaining continuity was feasible for school-age children for only four weeks. There is a need for long term mHealth use assessments among adolescents, to assess if the improvements in self-management are sustained (Alquran et al., 2018). Furthermore, use adherence decreases over time with the mHealth app, we think "booster" sessions may be necessary to address emerging issues in middle and late adolescence.

Evidence-based intervention

For an intervention to be effective and successful, its design must be informed using evidence-based theoretical approaches, clinical practice guidelines, or skill-based approaches (Stiles-Shields et al., 2019). The Life Course Model for SB that emphasizes that self-management should be established for individuals during childhood, especially during the school-age stage because of cognitive and motor development competencies evident during this time (Swanson, 2010). Furthermore, monitoring of the development of this self-management skill is especially important for children with SB (Swanson, 2010).

Our 2-step self-management intervention was developed by applying this Life Course Model and IFSMT. Based on our study, we found that the Life Course Model and IFSMT were useful frameworks for organizing the intervention, especially for the measures chosen to measure outcomes based on the IFSMT concepts. This theoretical framework guided the development of the self-management intervention used in this feasibility study.

All the contents and methods of the interventions were evaluated by a panel of eight experts, who have all been directly involved in or have expertise in caring for children with SB and who could determine the

Table 3
Feasibility comments for the 2-step self-management program by children with SB and their parents.

Category	children with SB (n = 5)	Parents (n = 5)
IEP	- "It was fun to get close to friends with SB and experience the laboratory exercise." - "It was good to play board game in the talk! talk! talk! time and I could know what I should do in such situation in detail." - "I could know about SB that I have, through this opportunity."	- "She was happy to meet friends with SB like her, and it was the opportunity to share her experience with them." - "She came to accept herself as she was and she thought positively about herself after the IEP." - "He has more confidence in himself than before." - "He seems to carry out self-management more than ever before with interest."
mHealth program	- "I can see what I'm doing about self-management, which helps me manage myself." - "Self-managing records let me know more about myself." - "It was good to know the amount of water intake, the time of voiding or CIC."	- "It helped her to recognize her lack of self-management." - "She stared to do the CIC herself with it." - "Through the mHealth program, he began to remember and care about his own water intake, amount of voiding and defecation."
Need to be complemented	- "I wish there was a date on the Starboard in the mobile app. That way, I think I'll know if I've identified and recorded all the categories of my self-management." - "The mobile app should add the function to note users whether they have recorded all of their own self-managing categories. There's no function to tell if you've made all the records."	- "For girls, I'd like to add the function which is a record of their menstruation to the app. Then they'll be able to see physiological changes during the menstrual period." - "I wish I could print out the records of my child's self-management (i.e., records the amount of water intake, voiding and defecation). I think it will be nice to take it with us to the hospital." - "I wish the mobile app had the ability to write special note. For example, if my child did a catheterization by himself and some blood becomes mixed with the urine… then it would be better to write such situation through text."

SB = spina bifida, IEP = Integrated Education Program, CIC = clean intermittent catheterization.

Table 4
Feasibility and user satisfaction evaluation of mHealth program by children with spina bifida (N = 5).

Dimension/item	Mean ± SD[a]	Min	Max	Range
System quality	27.2 ± 5.67	19	35	7–35
1. The mobile app works fine without errors	4.4 ± 0.89	3	5	1–5
2. The mobile app is easy to use	4.0 ± 0.71	3	5	1–5
3. The mobile app is-user-friendly	4.2 ± 0.84	3	5	1–5
4. The mobile app provides interactive features between user and app	3.6 ± 0.89	3	5	1–5
5. The mobile app provides a personalized information presentation	3.6 ± 1.14	2	5	1–5
6. The mobile app has attractive features to appeal to the users	3.4 ± 0.89	3	5	1–5
7. The mobile app provides high-speed information access	4.0 ± 1.22	2	5	1–5
Service quality	21.6 ± 2.07	18	23	5–25
1. The mobile app provides a proper level of explanation and assistance for using the app	4.0 ± 0.71	3	5	1–5
2. The mobile app department interacts extensively with users during the development of the mobile app	3.8 ± 0.84	3	5	1–5
3. The mobile app department provides high availability for consultation	4.6 ± 0.55	4	5	1–5
4. The mobile app department responds in a cooperative manner to user's suggestion for future	4.6 ± 0.55	4	5	1–5
5. The mobile app provides satisfactory technical support to users using the mobile app	4.6 ± 0.55	4	5	1–5
User satisfaction	13.4 ± 1.14	12	15	3–15
1. Most of the users bring a positive attitude or evaluation toward the mobile app function	4.4 ± 0.55	4	5	1–5
2. You think that the perceived utility about the mobile app is high	4.4 ± 0.55	4	5	1–5
3. You are satisfied with the mobile app	4.6 ± 0.55	4	5	1–5
Actual usage	11.8 ± 0.84	11	13	3–15
1. The frequency of use with the mobile app is high	4.4 ± 0.55	4	5	1–5
2. The mobile app is voluntary	4.0 ± 0.71	3	5	1–5
3. You depend upon the mobile app	3.4 ± 0.55	3	4	1–5
Total feasibility evaluation of mHealth program	74.0 ± 8.97	60	85	18–90

Note:
[a] Likert scale of 1–5 (with 1 indicating strongly disagree, 3 neutral and 5 strongly agree). SD = standard deviation.

suitability of the intervention. Although the outcomes were not statistically significant, participants experienced an understanding of their condition, and realized they shared common concerns with others with SB. They demonstrated motivation for self-management and feasibility for self-management maintenance by using mHealth independently.

User-centered intervention

Feedback was elicited during the evaluation stage as to whether the user-centered design had been successfully implemented, with the feasibility intervention survey for children with SB and their parents. Their comments on the feasibility and user satisfaction were fully reflected in the revisions and improvement. We modified both the onsite integrated education program and mHealth program to satisfy their requirements, which elicited favorable user responses.

Effectiveness of intervention

Although the 2-step self-management developed in this study was a valuable intervention for children with SB, it was difficult to determine the statistical effectiveness of variables before and after intervention because our sample included only 5 participants. However, the self-management behavior decreased immediately after the onsite IEP in three participants. We surmise that the reason for this may be because parental expectation was increased during the onsite IEP, as parents could also learn about IEP through the uploaded presentation files on the mHealth app. Initially, based on the IFSMT (Ryan & Sawin, 2009), we expected that contextual factors such as school adjustment, and process factors such as self-management knowledge and self-efficacy could be improved by this 2-step intervention. This affected the self-management behavior and quality of life of children with SB. Despite the results and our small sample, comments of children with SB and their parents were very positive, and reflected motivated self-management behavior after onsite IEP, with reported sustainability of their self-management behavior. Testing of this intervention model in other studies with larger samples is warranted.

Limitations

This study had several limitations. The sample size was small, and the study lacked a control group. We explored the feasibility of this program that combined two types of interventions and mHealth for school-aged children with SB with a small sample. However, now that this study has identified the feasibility and the functionality of this intervention, further studies are needed for testing that include larger samples, a control group and more in-depth statistical analysis of participants' data. Moreover, designing and developing a generalizable and fully accessible mHealth program should be considered in the future to improve the accessibility of these mHealth tools.

Table 5
Effects of the 2-step self-management program in children with spina bifida at follow-up (N = 5).

	Mean ± SD			*P*-value			
	Pre	Post 1	Post 2	Overall[a]	Pre-Post 1[b]	Pre-Post 2[b]	Post 1-Post 2[b]
Children with SB							
School adjustment	91.6 ± 12.9	94.4 ± 14.0	93.8 ± 11.0	0.408	0.176	0.581	0.684
Self-care knowledge	10.2 ± 2.0	11.6 ± 2.7	11.4 ± 2.9	0.106	0.102	0.141	0.317
Self-efficacy	29.4 ± 4.7	30.8 ± 5.9	31.2 ± 6.1	0.362	0.223	0.416	0.683
Parents of children with SB							
Child's self-care behavior	37.2 ± 7.6	34.2 ± 7.3	40.6 ± 3.8	0.056	0.416	0.176	**0.043**
Quality of life of the children	170.0 ± 20.4	179.2 ± 12.2	182.4 ± 15.9	0.097	0.223	0.068	0.225

SD = standard deviation, SB = spina bifida. Bold emphasis statistically significance at $P<.05$.
[a] Repeated measures ANOVA.
[b] Wilcoxon signed-rank test.

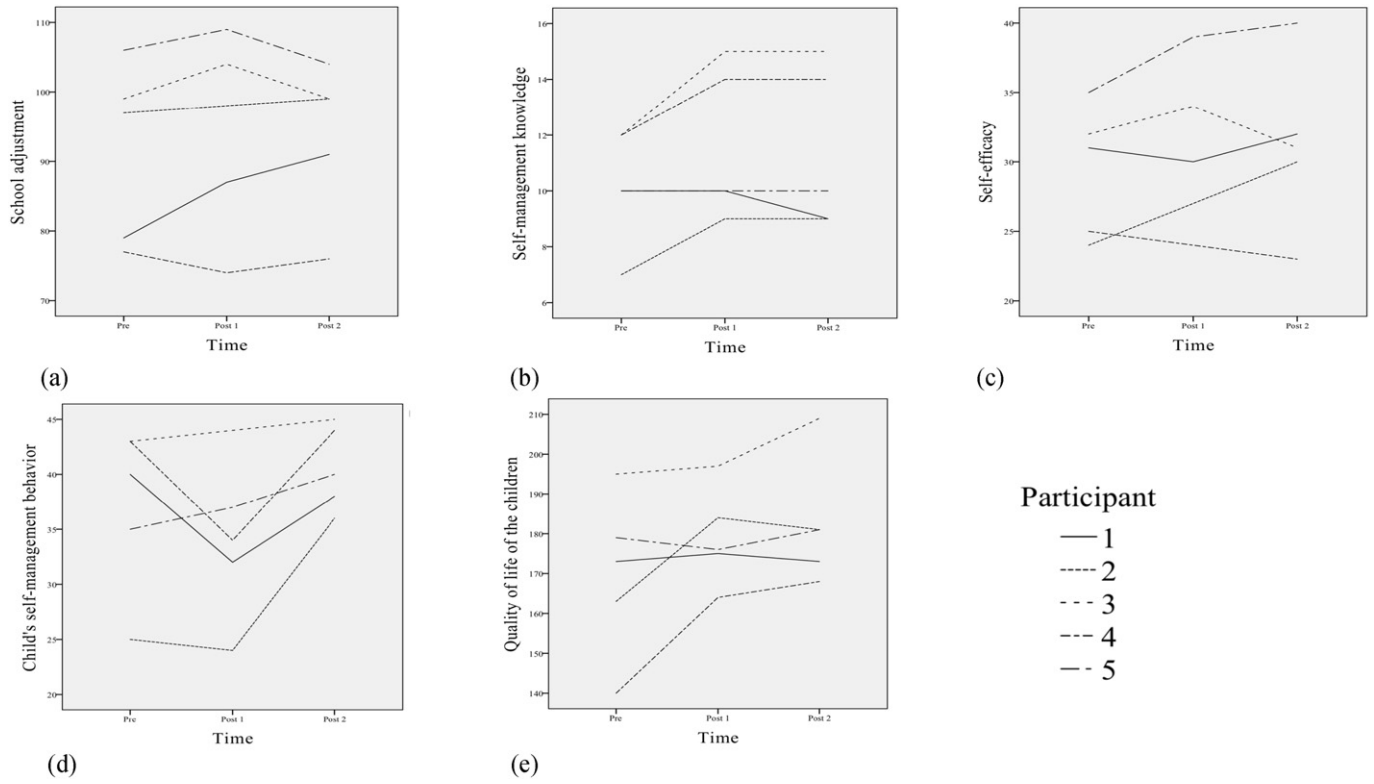

Fig. 3. Effects of the 2-step self-management program (N = 5). (a) School adjustment, (b) self-management knowledge, (c) self-efficacy, (d) children's self-management behavior, and (e) quality of life of the children.

Additionally, there are limitations associated with the measurements used for this study. While the Parkin's health-related quality of life tool has acceptable internal reliability, it also has multiple items with ceiling effects, has no evidence of any factor structure or validity and is not a strong HRQOL instrument (Sawin & Bellin, 2010). Until recently, there was not a suitable age- and condition-specific alternative to Parkin's in Korea; however, a new qulity of life (QOL) tool for children with SB has been developed in English (Szymanski et al., 2016). Therefore, future studies could take advantage of transcultural validation research to improve these measurement limitations.

Clinical implications

Health care providers are responsible for empowering children with SB to develop self-management skills. The findings from this study suggest that a 2-step self-management program combining onsite integrative education and mHealth program provided by pediatric nurses can be used effectively for school-aged children. Integrated education programs enable children with SB to understand their condition and to demonstrate and practice self-management themselves with the help of pediatric nurses. Utilizing mHealth assists children with convenient access to maintain their self-management. Therefore, in line with the conventions of pediatric nursing interventions, this innovative intervention program could be adapted for children with chronic conditions, with positive effects on their self-management. Furthermore, we recommend that health care providers enable the use of children's self-management records on this app, to share their adherence information. To achieve this, we would need to create an additional function in mHealth, which conveys information about the child's record to the electronic health record, in the hospital setting. Ultimately this function could provide tailored monitoring of children with SB in order to promote their self-management adherence.

Conclusion

Children with SB encounter various challenges and difficulties. In particular, school-aged children experience many barriers to self-management in the school environment. Achieving self-management at the school-age stage is important in maintaining health and quality of life into adulthood. This innovative 2-step self-management intervention program avoids the disadvantages of existing single interventions, and this study support the possibility of mHealth technology interventions for children with SB.

CRediT authorship contribution statement

Eun Kyoung Choi: Conceptualization, Data curation, Formal analysis, Funding acquisition, Investigation, Methodology, Project administration, Supervision, Validation, Writing -original draft, Writing – review & editing. **Eunyoung Jung:** Data curation, Investigation, Methodology, Validation, Writing - review & editing. **Yoonhye Ji:** Conceptualization, Data curation, Investigation, Methodology, Validation, Writing - review & editing. **Eunjeong Bae:** Conceptualization, Data curation, Formal analysis, Investigation, Methodology, Resources, Software, Validation, Writing -original draft, Writing – review & editing.

Declaration of competing interest

None.

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