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Parents' Needs Concerning Their Children with Spina Bifida in South Korea: A Mixed Method Study

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ABSTRACT

Purpose: This study aimed to determine the needs of children with spina bifida (SB) and their families from their parents' perspective in South Korea.
Design and methods: This was a convergent mixed methods study design. From December 2016 to February 2017, parents of children with SB participated in a quantitative prospective observational study (N = 164), using the Family Needs Assessment Tool. Qualitative focus group interviews were conducted, according to three developmental stages (N = 15) in May 2017. Integrated analyses were conducted jointly by merging the quantitative and qualitative findings.
Results: Quantitative findings revealed very high parental needs in three assessment domains: information, healthcare service/program, and difficulties related to healthcare. Ten qualitative themes were identified in these 3 domains. Quantitative and qualitative methods enabled more extensive findings. Comparison and merging of the data resulted in six confirmed and four expanded findings. In particular, we identified the need for a child-focused self-management program, a bladder/bowel disability awareness program, welfare policies, and partnership with healthcare professionals as the expanded findings.
Conclusion: This mixed method study provided empirical evidence to help better understand the complex needs of parents of children with SB.
Practice implications: When developing and providing healthcare education and service to families of children with SB, especially, in countries where SB educational programs have not been established yet, it is important to develop them based on their own needs, which may vary based on the child's developmental stage and socio-cultural characteristics.

Introduction

Spina bifida (SB) is the most common type of neural tube defect (NTD) that affects the central nervous system (Copp et al., 2015). SB occurs when the spinal cord does not develop properly due to incomplete closure of the neural tube at approximately 28 days of gestation (Phillips, Burton, & Evans, 2017). In the United States, approximately 3.61 out of 10,000 newborns are born with SB each year (International Clearinghouse for Birth Defects Surveillance and Research, 2008). In Korea, the incidence is similar to that seen in England and Canada, with approximately 2.62 newborns with SB per 10,000 live births (Choi et al., 2009).

Due to improved management for children with SB over the past five decades, more adults are now living with SB (Szymanski, Cain, Hardacker, & Misseri, 2017). However, there is growing evidence that SB can negatively affect not only the children with SB, but also their families (Rofail, Maguire, Kissner, Colligs, & Abetz-Webb, 2013). Children with SB can have impairments and disabilities associated with spinal cord injury, as well as bladder and bowel incontinence and orthopedic deformities. These issues involve cognitive impairment, poor academic performance, poor emotional functioning, mental health issues, low self-esteem and negative self-image challenges, and difficulties with social functioning and relationships (Bakaniene, Prasauskiene, & Vaiciene-

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Magistris, 2016; Rofail et al., 2013). Furthermore, families and caregivers are affected in terms of their activities of daily living, work, use of time, and parental responsibilities (Chaplin, Schweitzer, & Perkoulidis, 2005). Furthermore, they have negative issues concerning confidence, emotions, mental health, stress, social effects, psychosocial adjustment, relationships within the family, social support, and coping strategies (Rofail et al., 2013).

The difficulties faced by children with SB and their families are common, but the degree of difficulties is different between Western and Eastern countries depending on cultural characteristics. One reason for the differences that exacerbates the difficulties in Eastern countries is Confucianism. Confucianism is associated with disease-related stigma because being or having a child who is different is implied as being inferior to healthy people (Choi, & Yoo, 2015). As the Korean society has a very homogenous group of people (Choi, & Yoo, 2015), this negative attitude towards differences might be stronger in this setting. Additionally, it could continue due to the lack of opportunities to understand people with SB and cause more difficulties in children with SB and their families.

The level of intervention for children and families with SB also differs between the Western and the Eastern countries. The Centers for Disease Control and Prevention (CDC) established a National Spina Bifida Patient Registry (NSBPR) to facilitate research and to improve clinical care for children and adults with SB in the United States in 2008 (Sawin et al., 2015). Many healthcare providers and researchers have been working to reduce the negative effects of SB and increase normalization for children with SB and for their families. Consequently, since 2000, various interventions have been developed for people with SB and their families, such as fetal surgery for a fetus identified as having SB, and the development of a transition clinic for adolescents and adults with SB (Blumenfeld & Belfort, 2018; Fremion, Morrison-Jacobus, Castillo, Castillo, & Ostermaier, 2017; Hettel, Tran, Szymanski, Misseri, & Wood, 2018; Joyeux et al., 2018). However, more countries in the East have provided advanced medical treatment, many intervention programs have focused mainly on medical needs and not on family focused needs. There has been very limited research concerning the perspective of people with SB and their families, such as identifying what specific services families require from healthcare providers (Buran, McDaniel, & Brei, 2002; Groisman et al., 2013; Kennedy et al., 1998). These studies have been quantitative in design and have lacked a qualitative perspective.

Furthermore, cultural differences are very important when the needs of people with chronic conditions are being determined because they affect policies, programs, services, and interventions within countries or cultures (Groisman et al., 2013). In most Asian countries, a systematic patient registry for this rare disease is not available, with very few studies identifying the needs of people with SB. The needs of parents of children with SB are likely to differ in Western countries; for example, factors such as insurance systems, welfare services, and social stigma may differ. Therefore, it is important to understand cultural backgrounds and the healthcare system when assessing the needs of children with SB. From this perspective, the mixed method study could identify parents' needs in caring for their children with SB through a determination of parents' perspectives and experiences.

Purpose

The aim of this study was to determine the needs of children with SB and their families from their parents' perspective in South Korea.

Design and methods

This study was a mixed method study using a convergent design. Using a convergent design, data are collected, with quantitative and qualitative data analyzed separately and examined to determine to what extent quantitative results are confirmed through qualitative results after the results have been merged (Creswell & Plano Clark, 2018). Through conducting a cross-sectional questionnaire survey to investigate the needs of parents of children with SB, we identified their needs in depth and in greater detail through focus group interviews. We then integrated the merged results and the differences through comparing and analyzing the quantitative and qualitative results.

Participants

Quantitative study

Based on our selection criteria, we included parents of children <18 years old with SB without other disabilities. The participant parents were able to communicate and respond to the questionnaires and voluntarily consented to participate in the study. Participants included parents of children with SB who had registered at the SB clinic of a children's hospital in Seoul, which has the largest SB clinic in South Korea. In total, 164 questionnaires were included in the analysis. To determine the appropriate sample size for this study, a power analysis was performed using G*power 3.1. 9.2. (Universität Düsseldorf, German). In total, 156–171 participants were needed for a correlation analysis with a 0.05 α-level, 80% power, 0.25 median effect size, and 10% dropout rate; therefore, our sample size of 164 was sufficient.

Qualitative study

For the qualitative phase, among parents of children with SB who participated in the survey, those who had agreed to participate in a focus group interview were selected. Additionally, we used characteristics such as the age and sex of their children and their residing area to group participants after recruitment. This information was obtained to include participants from varying backgrounds within similar groups, if possible. According to a child's developmental stage, parents were divided into three groups (≤6, 7–12, and ≥13 years old) and each group consisted of between 4 and 6 participants, with a total of 15 participants.

Measures

Quantitative study

A Family Needs Assessment Tool (FNAT) (Rawlins, Rawlins, & Horner, 1990) has been developed to assess the needs of parents of children with chronic illness. A version of FNAT had been translated by a committee approach (Brislin, 1970). All questionnaire items were translated into Korean with the agreement of three bilingual nursing professors. Then, the content validation of the tool was undertaken by five expert researchers or clinicians with experience in the treatment of children with SB. As a result of the expert validity assessment, the “out of home placement” item for service/program needs was reduced to Content Validity Index (CVI) = 0.4, and it was deleted. The item “appropriate educational opportunities for children” for service/program needs was reduced to CVI = 0.6, which implied it was unclear, so this item was further modified to “appropriate educational opportunities for children (public education, private education)”. The other items were CVI = 0.8 or higher. The final FNAT included 53 items and three subscales: service/program needs (13 items), information needs (28 items), and difficulties related to healthcare (12 items). The service/program and information needs subscales had three response options, as follows: ‘Yes’ (you/your child has this need), ‘No’ (this is not a need for you/your child) and ‘Need Met’ (you/your child had this need but it has been met). Difficulties relating to the healthcare subscale provided three possible response options, as follows: rarely, sometimes, and frequently. Internal consistency results for the three subscales showed: service/ program, Cronbach's $\alpha = 0.856$; information needs, Cronbach's $\alpha = 0.947$; and difficulties related to healthcare, Cronbach's $\alpha = 0.780$.

Qualitative study

Focus group interviews were conducted to obtain further in-depth information. Three open-ended interview questions were developed and used to collect qualitative responses, as follows: “What services or programs do you need to solve difficulties or reduce difficulties experienced in raising a child with SB?”, “What information or training do you need as you raise your child with SB?”, and “What difficulties have you experienced in relation to medical services and how do you want to solve your difficulties?”

Data collection

Quantitative study

Two methods were used for data collection, namely, an face-to-face survey and an online survey. Data collection was conducted between December 2016 and February 2017. To conduct the face-to-face survey, we first screened the participants, with the cooperation of a pediatric urologist and an advanced nurse practitioner who worked at the SB clinic at Children's Hospital, Seoul, Korea. Parents of children with SB who had consented to participate voluntarily in the study completed the questionnaire on their own. The survey was provided using trained research assistants and was conducted at an external location for participants.

Study participants were required to complete the FNAT using an online survey on an online parental self-support community website, namely, the Korea Spina Bifida Patient Association in South Korea. This group had formed approximately 15 years prior to the survey and has served as a peer support group for parents of children with SB. The group is not owned by a commercial or professional entity but is maintained and moderated by approximately six parent volunteers, and is the only community group for parents of children with SB in South Korea. The survey was hosted on this online community website. Potential participants were provided with a link to the project information sheet. After reading the information sheet, interested participants were then able to access the online survey (hosted by Google Forms). Details of the study were provided and written informed consent was obtained online.

Upon completion of the face-to-face and online surveys, a token gift of US $10.00 was offered to the participating parents.

Qualitative study

The focus groups were classified into age groups, and focus group interviews were undertaken with mothers of infants, toddlers and preschool children, elementary school children, and adolescents with SB. Focus group interviews were held in a quiet meeting room at the College of Nursing on different dates in May 2017. Initially, the moderator provided a brief introduction of the purpose and progress of this study. The focus group interviews were both audio-recorded and transcribed verbatim by a trained research assistant. The interview time for each group was between 2 h and 2.5 h. One researcher with experience facilitating focus group interviews served as the moderator. Following the interview, a token gift of US $45.00 was offered to participating parents.

Ethical considerations

Ethical approval for this study was obtained from the Institutional Review Board of the [Yonsei] University Health System. The researchers obtained written informed consent from participating parents after explaining the purpose and method of this study. Once written informed consent had been obtained, a unique study identifier number was assigned to ensure confidentiality and anonymity. All participants had the opportunity to decline participation at any point. In terms of focus group interviews, participants were assured that the interviewers were independent of the healthcare services they received, and that the interviews would be confidential. After explaining to the participants that the interview would be recorded, and the recorded content would be protected and remain confidential, participant permission to proceed was obtained.

Analysis

Quantitative study

SPSS version 24.0 (IBM, Seoul, South Korea) was used to analyze descriptive statistics such as frequency, percentage, mean, and standard deviation (SD) of the survey data.

Qualitative study

The analysis was conducted using content analysis in which data were coded and analytic themes developed. First, the focus group verbatim transcripts were read several times to gain familiarity with the data. Then, two researchers (Choi, EK and Jang, M) separately and systematically analyzed the data from the focus group transcripts using coding guidelines. The two researchers reached a consensus concerning the coding process, and the final coding result from this process was then used to combine the codes into themes.

Integration of quantitative and qualitative data

For the mixed method analysis, qualitative and the quantitative results were compared side by side (Creswell & Plano Clark, 2018) to confirm quantitative results where possible using qualitative outcomes and identify differences in quantitative and qualitative outcomes.

Results

Quantitative results

The study respondents comprised 164 parents of children with SB, of whom 82.8% were mothers. The mean age of the respondents was 39.99 ± 5.54 years, 66.8% of respondents had graduated from college, and 92.7% reported their economic status as middle class. Among the children, 95 (57.9%) were boys, and the mean age of the children was 7.83 ± 5.33 years. Of these, 15 (9.3%) had a shunt to control hydrocephalus, and 27.7% of the children needed aids for mobility. Regarding the method of bladder emptying, 47.7% of the children were in need of clean intermittent catheterization (CIC) and 21.4% were wearing pads due to urinary incontinence. For the method of bowel emptying, 54.2% of the children needed additional interventions to defecate regularly, including digital stimulator/manual extraction, laxatives, transanal irrigation, and antegrade continence enemas (Table 1), and 66 (42.9%) children had experience of fecal incontinence.

The area of greatest need was in terms of information as determined by the FNAT. The mean number of items parents indicated on the 28-item information needs subscale was 20.56 (possible range, 0–28), whereas the mean number of items parents indicated on the 13-item service/program needs scale was 8.27 (possible range, 0–13), and the 12-item difficulties related to healthcare scale was 17.48 (possible range, 12–30). In terms of information needs, all items within the information needs subscale were endorsed by >50% of the study sample, and 14 of 28 items were needed from >75% of parents (Fig. 1). In terms of service/program needs as determined by the FNAT, 12 of 14 items were needed from 50% of parents (Fig. 2). A smaller proportion of the study sample endorsed items comprising the difficulties related to healthcare subscale as determined by the FNAT; only four items were noted as areas of need by >10% of the respondents (Fig. 3).

Qualitative results

Based on qualitative data analysis, the following 10 themes were identified in 3 domains, namely, needs assessment on information, healthcare services, and obstacles encountered and the desired support,

Table 1
Characteristics of the respondents and of children with spina bifida (N = 164).

Variable	Category	N (%)
Respondents		
Sex[a]	Male	28 (17.2)
	Female	135 (82.8)
Age (years), mean ± SD (min, max)[a]		39.99 ± 5.54 (27, 58)
Highest level of education[a]	High school	54 (33.2)
	College or university	92 (56.4)
	Graduate school	17 (10.4)
Perceived level of economic status[a]	High	1 (0.6)
	Upper middle	18 (11.1)
	Middle	93 (57.1)
	Lower middle	40 (24.5)
	Low	11 (6.7)
Children with spina bifida		
Sex	Male	95 (57.9)
	Female	69 (42.1)
Age (years), mean ± SD (min, max)[a]		7.83 ± 5.33 (0, 18)
	≤6 years old	77(47.0)
	7–12 years old	50(30.4)
	≥13 years old	37(22.6)
Age at time of SB diagnosis (months)[a], mean ± SD (min, max)		6.41 ± 15.36 (0, 78)
Age at time of the NS operation (months)[a], mean ± SD (min, max)		9.08 ± 16.44 (0, 84)
VP shunt in situ[a]	Yes	15 (9.3)
	No	138 (85.7)
	Don't know	8 (5.0)
Mobility[a]	Use a wheelchair	4 (2.5)
	Use of aid	40 (25.2)
	No aid	115 (72.3)
Method of bladder emptying[b]	Self-voiding	62 (30.9)
	Clean intermittent catheterization	96 (47.7)
	Pad	43 (21.4)
Method of bowel emptying[b]	Straining/bearing down to empty	79 (45.8)
	Digital stimulator/manual extraction	13 (7.7)
	Medication	32 (18.6)
	Transanal irrigation	40 (23.4)
	Antegrade continence enema	8 (4.7)
Fecal incontinence[a]	Yes	66 (42.9)
	No	88 (57.1)

NS: neurosurgical, SB: spina bifida.
[a] Excluded non-responses.
[b] Multiple choice.

using a total of three focus group interviews involving 15 invited mothers.

Theme 1: information needs

Most participants were uncertain or fearful about whether SB would lead to other health issues. The participants stated that self-care education programs, such as instructions for clean intermittent catheterization (CIC) and enema, are needed before beginning elementary school, and sex education tailored to children with SB is needed in elementary, middle, and high school. Furthermore, they had concerns for their children's future and hoped that their children would overcome their challenges. The mothers stated that the opportunity to listen to the experiences of young adults with the same disease had a positive effect on their children.

Effects on physical growth and development

> "*I'm worried because I don't know much about the changes that occur after surgery. It was so difficult to get information. When we visited the hospital because of constipation, the doctors simply said it was because of the disease. It was just so frustrating*"
>
> [(preschooler's mom 4)]

Self-management program

> "*My child had to learn instructions for CIC before he/she goes to elementary school. I went to his/her school until he/she was in the 5th grade to do it for him/her because I thought he/she could not do it by him/herself. In hindsight, I think that it was not for the good of my child*".
>
> [(schooler's mom 5)]

> "*What parents can do for their child is to teach them how to do CIC, but I wish that there was a program that systematically teaches self-care*".
>
> [(schooler's mom 3)]

Sexuality & sexual education

> "*I think a stepwise sex education program is needed*".
>
> [(schooler's mom 2)]

> "*Sex education seems more difficult because of CIC and enema. It is also difficult for parents to teach their child about sex. Since my child goes to the hospital often, I think it would be helpful if the nurses could provide a sex education class.*"
>
> [(adolescent's mom 3)]

Planning for child's future

> "*Our children need to prepare for the future, but most of the time, they believe they can't do it because they're sick. There's no use in this case regardless of what a mother could say to them for encouragement*".
>
> [(schooler's mom 1)]

> "*I wish that we can frequently meet with adults who have SB and have successfully adjusted to society. There was an open lecture by a successful person with SB, and my child learned a lot from the lecture*".
>
> [(adolescent's mom 2)]

Theme 2: service needs

Parents of children with SB stated that they needed family counseling concerning the children's diagnosis and family adjustment, concerning child-raising practices that are appropriate for different developmental stages, and concerning sibling relationships. Particularly, parents with young children wanted childcare services for emergency situations, as it was usually the mothers who were solely in charge of caring for their children and performing CIC when their children were young. They considered that family difficulties could be resolved to a certain extent through self-help groups for parents. They stated that the social perception of bladder and bowel dysfunction needs to change in Korea, and that more welfare policies should be implemented.

Counseling services for the family

> "*I believe that a mother has to be ready in order to rear her children. I was not prepared at all, and I blamed myself for my child's disease. I*

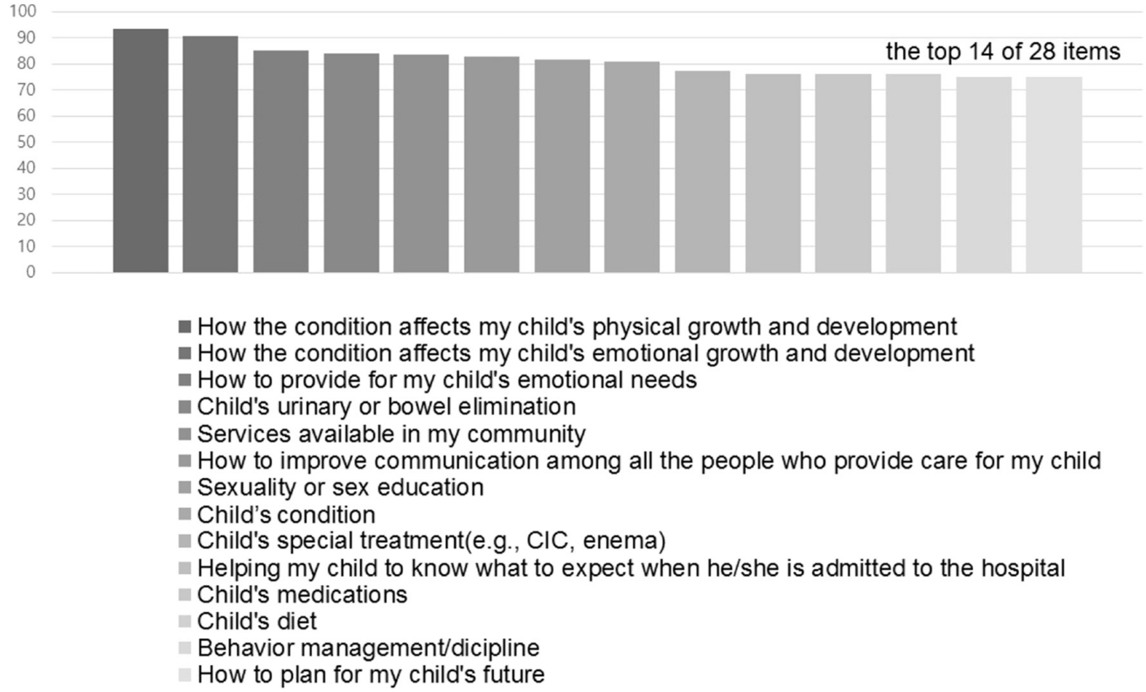

Fig. 1. Information needs from Family Needs Assessment Tool (FNAT).

know that these problems are caused by the disease, *but I just can't accept it in my heart. I don't know how to rear my child*; *I need help*".

[(preschooler's mom 3)]

"*I was shocked when my child was playing with dolls and was doing CIC on the doll. I know it's not wrong, but I was just so shocked and thought that my child needed psychological counseling. I don't know how to accept and rear him/her*".

[(schooler's mom 5)]

"*Family counseling is needed not only for us*, *who hide our children's disease behind our backs*, *but also for our other children who are not sick. My other child was jealous of his sick brother because all of the*

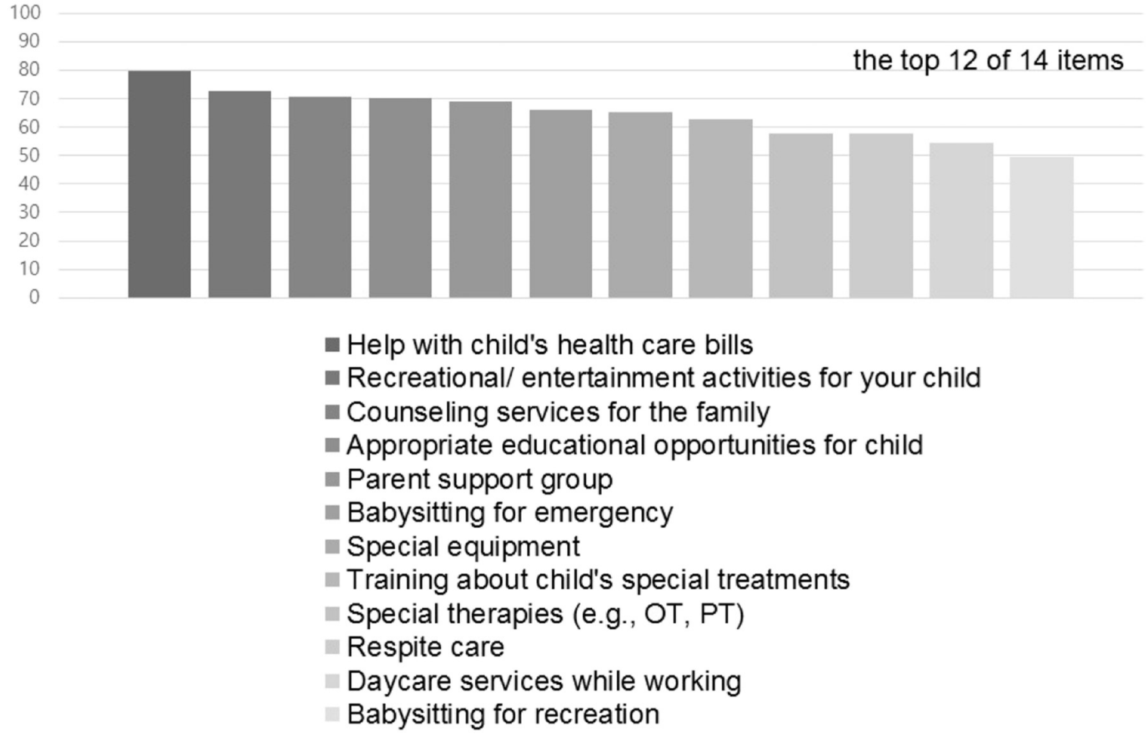

Fig. 2. Service/program needs from Family Needs Assessment Tool (FNAT).

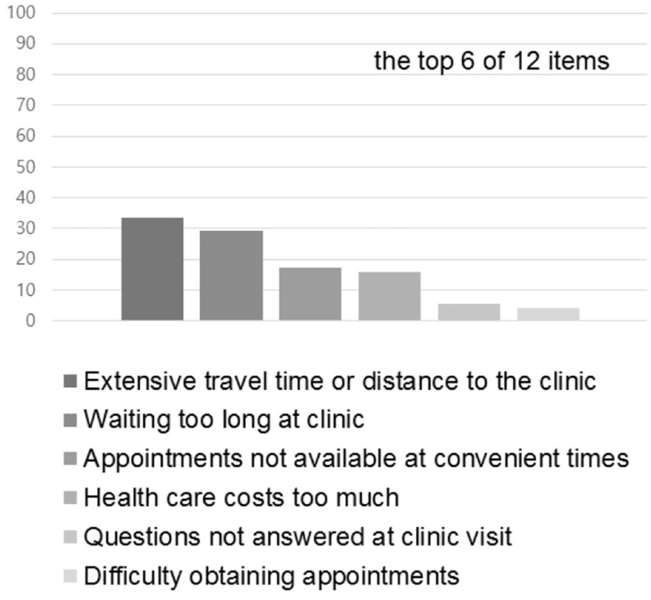

Fig. 3. Difficulties related to healthcare from Family Needs Assessment Tool (FNAT).

> *money was spent on him and only he gets to go to the hospital with his mom. I don't know how to treat my other child. This is something that I think about more often as my child grows older*".
>
> [(adolescent's mom 2)]

Babysitting service for emergency situations

> "*I have to be on standby 24/7 to perform CIC and enema for my child. I have no one to turn to when I have an emergency. My child doesn't qualify for the government-provided babysitter service for non-disabled children because of a health problem and also does not qualify for the babysitter service for disabled children because my child can't get disability registration. Our children are placed in this blind spot of welfare*".
>
> [(preschooler's mom 2)]

Parental support group

> "*In a sense, parents who have dealt with this problem longer are like my teachers. I wish there are programs that allow parents to share information regularly*".
>
> [(schooler's mom 3)]

> "*I wish that the* SB *Association founded by parents would have a stronger voice. It's been long since it was founded, but I'm sad that it's not as active as it should be. For it to be more active, large hospitals need to offer more help. Especially, parents of babies newly diagnosed with SB should be introduced to parent groups*".
>
> [(adolescent's mom 1)]

Improved bladder/bowel dysfunction awareness program for public, school teachers, and healthcare providers

> "*Our country is very lenient towards physical disabilities, such as people in wheelchairs, because it's visible, so they're more lenient, but people with invisible disabilities like us scares people. But Koreans are disgusted by bladder and bowel problems, and there are just so many people, like school teachers and even primary hospital staff, who are not aware about the existence of these health conditions*".
>
> [(adolescent's mom 2)]

> "*I have called the Ministry of Health and Welfare as well as the Office of Education regarding the need for school education programs to change the perception of bladder and bowel dysfunction. They just toss the ball around. They toss the responsibility to someone else and just say that unfortunately, there is nothing they can do. This is why our children can't get any help from schools*".
>
> [(schooler's mom 4)]

Welfare policies

> "*At first, going outside was just so stressful. I always thought, where should I go and what should I do? Once I go outside, I first check whether there is an area to change diapers. If there is no such area, I wouldn't go. But now, I often do CIC inside the car. There is a serious shortage of restrooms that are equipped with a changing station where I can lay down my child to perform CIC*".
>
> [(preschooler's 3)]

> "*Although the benefit extension policy is now in place and catheters for CIC are covered, the transanal irrigation set and catheters are not covered yet. My child doesn't qualify for disability benefits and can't get private insurance either. The burden for medical costs is just mounting as my child grows older*".
>
> [(adolescent's mom 4)]

Theme 3: obstacles encountered and the desired support

Many parents thought that they had not been given adequate information regarding their child's treatment plan because the outpatient clinic was too busy, and that treatment plans or other information frequently changed. Hence, they wanted medical staff to consult with them prior to deciding on their child's treatment plan.

Partnership with professionals

> "*I don't know if it's because my child's health state differs every time, but our doctor sometimes says one thing in one visit and another thing in the next visit, so I take notes every time and ask questions until my doubts are resolved*"
>
> [(schooler's mom 1)]

Inferences drawn

The themes emerging from the qualitative findings were compared and merged with the quantitative results in terms of parents' needs on information, healthcare services, and obstacles in caring for their children. Comparison and merging of the data resulted in confirmed and expanded findings (Table 2).

Regarding experiences of raising children with SB, the parents' needs differed on some specific items according to a child's stage of development. There were many requests for more information related to diseases and babysitting service in emergency situations at the preschool stage; whereas, several requests were made for sex education, and for future planning for children of school age and adolescence. Parents hoped that family support groups would become even more active and organized. Counseling services for the family (parents and siblings)

Table 2
The results of inferences.

Type of needs	Confirmation			Expansion
	Toddler & preschooler	School-age	Adolescents	All ages
Information	Effects on physical growth and development	Sexuality & Sex education Planning for child's future	Sexuality & Sexual education Planning for child's future	Self-management program for children
Services	Counseling services for the family Babysitting for emergency	Counseling services for the family Parental support group	Counseling services for the family Parental support group	Improved bladder/bowel dysfunction awareness program for public, school teachers, and health care providers Welfare policies
Obstacle				Partnership with professionals

were services that all parents wanted. These six themes confirmed the quantitative findings.

Additionally, the themes of many parents identified a strong need for a self-management program for children, a bladder/bowel disability awareness program for the general population, welfare policies for people with bladder/bowel issues, and partnership with professionals. These themes provided additional information beyond the qualitative findings.

Discussion

This mixed method study allowed researchers to attain a comprehensive understanding of what parents of children with SB require when raising their children. In Western countries, where guidelines and clinics for children with SB care are well established, very limited research has been published since a survey was undertaken in 1990 (Kennedy et al., 1998). However, in many countries where SB educational programs have not been established, our study findings suggest that research studies and education programs and policies need to be established. Furthermore, in the countries that developed these SB educational programs well, the needs of parents and their children with SB require regular investigation to better maintain and develop well-established educational programs.

Parents raising children with SB have needs that are not being met by health and social welfare systems from which they obtain services. The parents in this study identified the highest number of needs in the information domain, followed by healthcare service/program needs and difficulties related to healthcare. In particular, all 28 items of the information needs and 12 of 14 items of the healthcare service/program needs were needed in >50% of parents. The high demand for such information and services/programs was significantly different from findings reported in a United States study (Buran, Sawin, Grayson, & Criss, 2009), which measured the needs of parents of children with cerebral palsy using the same tools. In Buran et al.'s (2009) study, all items of the information needs subscale were endorsed by <18%, and all items of the service/program subscale were endorsed by <40%. The difference in the needs of the parents of children is likely to be due to differences in disease-specific education programs and specialized clinics between South Korea and the United States. In countries where health and social welfare systems for people with rare diseases are not well established, as the results of this study reveal, the need for direct information regarding the physical and psychological effects of the disease on the child and disease-related treatment is higher than the demand for service domains such as economic, leisure, and family support. Therefore, when planning health and social welfare systems, it is necessary to consider priorities according to the culture of different countries.

Across all ages, the parents reported the need for self-management programs for children. According to the Life Course Model for SB (Swanson, 2010), self-management should be started with body awareness at preschool age, and a continued understanding of any impairment and sharing their condition with others should be encouraged and managed at school age. Then, children could take a lead on managing primary/secondary conditions during their adolescence. Specifically, health professionals expect self-management behaviors such as CIC, assisting the parent(s) with completing the bowel program, cleaning up after a bowel or bladder accident, and recognizing the signs of bowel problems and urinary tract infections in school-aged children with moderate-to-severe SB (Greenley, 2010). Additionally, parents in our study also asked for a family counseling program for improved communication within families and with other people, a sex education program for their children, and help with preparation for their child's future such as job training counseling. These identified needs coincided with those reported in the Life-Course Model for SB in the United States (Swanson, 2010). The Life-Course model was designed to address three broad functional domains: health and condition self-management; social and personal relationships; and education or income support (Thibadeau, Alriksson-Schmidt, & Zabel, 2010). Therefore, when planning an education program or healthcare services for children with SB and their families, a program with long-term perspectives based on the child's development cycle should be planned, similar to the Life-Course model for SB, and not be a one-off education program.

Parents of all developmental-age children strongly requested an improved bladder/bowel dysfunction awareness program for the public, school teachers, and healthcare providers. They viewed healthcare professionals (even many medical professionals) as having only limited and vague knowledge and understanding of SB. Therefore, parents only access tertiary children's hospital providers instead of primary care providers, even for simple health issues. The parents also had ongoing concerns that general education teachers (and even some special education teachers) had very little knowledge of the overall challenges facing the children with SB. These results were very similar to those of a study conducted in the United States (Kennedy et al., 1998). Therefore, when planning an improved rare diseases awareness program, systematic strategies are needed to provide regularly updated information on SB that addresses both the current issues and long-term expectations, not only for the public but also for school teachers and healthcare professionals.

Furthermore, parents also claimed that improvements to welfare policies relating to SB and/or bladder/bowel dysfunction are necessary. Many children with SB require CIC and transanal irrigation (Choi, Van Riper, Jang, & Han, 2018). Most Western countries cover the long-term costs of urinary and transanal irrigation catheters or their accessories. The Korean government recently amended a law to cover the costs of intermittent catheters to $8 per day, which has helped families afford up to six hydrophilic catheters for children with SB since 2013 (Lim, Lee, Davis, & Park, 2016). However, the catheter and accessories for transanal irrigation are not being covered by insurance companies, despite South Korea having a national health insurance system. Furthermore, because there is a lack of understanding in regard to bladder and bowel issues facing children with SB, only a limited number of restrooms are provided for people with disabilities in public spaces. The study findings reflect those of previous study findings that identified that the lack of a dedicated place for CIC was the most common challenge for school-aged children (Lim et al., 2016). Therefore, this issue and bladder/bowel dysfunction should be addressed through policy improvements and in the development self-management education programs. To improve policy, a national SB patient registry should be instigated in Korea, as in Western countries (Thibadeau, 2017).

Implications for practice

The findings from this study provide us with information and awareness related to parental needs of children with SB, especially Eastern countries including South Korea. Our finding aligned with those of Western families with respect to the need for information, healthcare service/programs, and difficulties related to healthcare based on the child's developmental stage. However, there are limitations to accepting the needs of Western parents as they are. According to the cultural background, additional needs other than those previously reported in Western studies were noted. Therefore, when developing and providing healthcare education and service to families of children with SB, especially, in countries where SB educational programs have not been established yet, it is important to develop them based on their own needs, which may vary based on the child's developmental stage and socio-cultural characteristics.

Limitations

This study has some limitations. Although the survey sample obtained the required sample size, a convenience survey sample recruited through the Korean Spina Bifida Association may not represent the geographical or clinical characteristics of all children with SB in South Korea. Second, the responses obtained from this sample were reflective of the views of parents participating in the Korean Spina Bifida Association who may have had greater understanding or support than parents not participating in any parental self-help group. Finally, the generalizability of this study's results to fathers of children with SB might be limited due to the low number of fathers who participated in this study.

Conclusion

This study provided insights into the needs of parents of children with SB. The findings showed very high parental needs in relation to information and healthcare services. Additionally, qualitative findings supported our quantitative findings concerning the effects of SB on physical growth and development, sex education, planning for a child's future, counseling services for the family, babysitting services in emergency situations, and parental support groups. Qualitative findings identified further factors involving specific services and policies such as: a self-management program; an improved bladder/bowel dysfunction awareness program for the public, school teachers, and healthcare providers; welfare policies, and; improved partnership with healthcare professionals. Therefore, it is important to consider a child's developmental stage and socio-cultural characteristics when developing and providing healthcare services for children with SB.

CRediT authorship contribution statement

Eun Kyoung Choi: Assistant ProfessorConceptualization, Data curation, Formal analysis, Investigation, Methodology, Supervision, Validation, Writing - original draft, Writing - review & editing. **Yoonhye Ji:** Advanced Pediatric Nurse PractitionerConceptualization, Data curation, Investigation, Methodology, Validation, Writing - review & editing. **Eunjeong Bae:** Graduate studentData curation, Investigation, Methodology, Validation.Writing - review & editing. **Mina Jang:** Doctoral Student

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