

TRANSITION TO ADULTHOOD:  
EXPERIENCE OF YOUTH WITH SPINA BIFIDA IN JAPAN

by

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## ABSTRACT

### TRANSITION TO ADULTHOOD: EXPERIENCE OF YOUTH WITH SPINA BIFIDA IN JAPAN

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Under the Supervision of Professor Dr. Jennifer J. Doering

**Purpose:** The purpose of this dissertation was to describe the experiences of transition to adulthood for emerging adults with spina bifida (SB) in Japan.

**Background:** Spina bifida is one of the most common congenital conditions that affect functions of multiple body systems, including orthopedic, urologic, gastrointestinal, neurological, and cognitive. The prevalence of SB in Japan was 5.18 cases per 10,000 in 2012 and the long-term trends of the prevalence did not decline despite the recommendation of folic acid intake by the Japanese Government in 2000. Over 70% of individuals with this condition can survive into adulthood today because of the advancement of neurological management. As a result, transition to adulthood became one of the most significant issues for individuals with SB and their families in Japan. However, experiences during transition for emerging adults with SB in Japan remains unknown.

**Methods:** Qualitative descriptive design was used to explore experiences of the emerging adults with SB during transition to adulthood. Semi-structured telephone or Skype interviews were conducted with eight emerging adults with SB aged between 20 and 29 years living in Japan.

**Results:** Six core themes were identified: (a) struggling, (b) finding the meaning of “being an adult”, (c) learning to implement self-management, (d) health care, (e) processes and challenges in establishing and retaining relationship with friends, and (f) broadening scope of experiences. The findings highlighted participants’ experiences of struggling during transition to adulthood. In particular, participants had shared experiences of struggling to find a job and with working. Some participants also described their experiences of transferring to adult health care without planning or preparation.

**Discussion:** Three manuscripts were generated in this dissertation: two syntheses of literature written in English and published in Japanese journals, respectively, and a qualitative descriptive study. In the qualitative findings, participants’ experiences of struggling during transition to adulthood were persistent and sustained, especially when trying to find a job and making the transition to adult health care. Health and social programs assisting emerging adults with SB in achieving milestones for adulthood and transitioning to adult health care in Japanese social and health care context are needed.

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## DEDICATION

To

My husband Ryosuke,

My sons Ken and Shuji,

My parents Masashi and Yoko,

And my sister Misa,

I could not have completed this dissertation without your support. Thank you.

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## CHAPTER ONE

Spina bifida (SB) is one of the most common congenital conditions that affect functions of multiple body systems, including physical, neurological, and cognitive. Although the prevalence of SB has generally been declining worldwide, in Japan there was an increase in the prevalence between 1980 and 2000 (Takimoto & Tamura, 2006). Between 1997 and 2005, the overall prevalence of SB in Japan was 4.6 cases per 10,000 (Hirahara & Tanaka, 2007). More recently, the prevalence of SB in Japan was 5.18 cases per 10,000 in 2012 (International Clearinghouse for Birth Defects Surveillance and Research, 2014), while it was approximately 6.0 in U.K. (Morris et al., 2016), 3.31 in Canada and 4.69 in Western Australia (International Clearinghouse for Birth Defects Surveillance and Research, 2014). The long-term trends of prevalence of neural tube defects did not decline despite the recommendation of supplement intake by the Japanese Government in 2000 (Yorifuji, 2019).

The prognosis of the individual with SB has been greatly improved in the last 50 years. The survival rate of children was approximately 17% in 1950s -1960s (Laurence, 1974). Today, over 70% of children with SB can reach adulthood because neurosurgical management has markedly improved (Bowman et al., 2001). Laurence (1974) reported that the possibility for long-term survival for these children increased after the ventricular cerebrospinal fluid shunt surgery was introduced. On the other hand, Oakeshott, Hunt, Poulton, and Reid (2009) found that the mortality rate between the ages of 5 and 40 was still ten times higher than the national average for the cohort with myelomeningocele born and treated in a hospital in UK between 1963 and 1971, which questions improved life expectancy in individuals with SB. However, with advancement in medical management of the condition, it can be safely assumed that the

possibility of entry into adult life for the individuals with SB is increasing both worldwide and in Japan.

As a result of the improved prognosis, Japanese adolescents and emerging adults (AEA) with SB and their families are confronting various new challenges related to transition to adulthood (Date & Ito, 2003; Koike, 2011). The AEA and their families have concerns about education, employment, and familial issues in addition to concerns on management of the condition (Date & Ito, 2003), which are all related to transitions to adult life and to adult health care. It has also been found that individuals with SB become aware of their difference from others in the process of developing self-management behaviors and struggle to cope with the difficulties related to the condition and maintenance of their self-esteem (Koike, 2008, 2011). Thus, transition to adulthood is currently one of the most significant issues for individuals with SB and their families in Japan.

### **Significance**

Previous research conducted in the Netherlands and the United States has shown that chronic conditions have negative impact on the achievement of developmental milestones critical to healthy transition into adulthood (Davis et al., 2006; Kinavey, 2007; Ridosh et al., 2011; Sawin et al., 2009; Stam et al., 2006). Stam, Hartman, Deurloo, Groothoff, and Grootenhuis (2006) revealed that emerging adults with chronic conditions in the Netherlands were delayed or had not achieved milestones in the domains including autonomy development and social development compared to healthy counterparts. These delays were also common in youth with SB in the United States. Although the development of some basic self-management behaviors (e.g., dressing appropriately, planning activities with peers, cooking pre-packaged meals) were

accomplished by adolescents with SB, many were delayed by 2-5 years and others typically achieved by adolescents' peers (e.g., managing an allowance, cooking independently, doing their own laundry) were not achieved at all in adolescents with SB (Davis et al., 2006). Studies conducted in the United States showed that AEA with SB also often confront psychosocial challenges, such as complex adjustment demands, psychological distress, and social isolation (Kinavey, 2007; Ridosh et al., 2011; Sawin et al., 2009) that may become barriers to healthy transition to adulthood. In a study from the adolescents' perspective, Sawin et al. (2009) reported that the parents delicately balanced their desire to protect their children and need for pushing toward independence.

Existing research findings in Japan, however, only provide limited knowledge with regard to transition for Japanese AEA although the literature is gradually growing. Existing research on the experience of individuals with SB is somewhat limited in scope (e.g., limited to school settings or limited to bowel and bladder management) (Koike, 2008, 2011). Research that specifically focuses on the experiences during adolescence and emerging adulthood in individuals with SB, including challenges the individuals are faced with as well as positive aspects during the transition to adulthood, is also extremely limited. Therefore, it is important to investigate the experiences of Japanese emerging adults with SB during their transition.

Due to the lack of empirical research in the area, qualitative research, whose purpose is to explore the meanings people construct and attach to phenomena using a naturalistic approach (Denzin & Lincoln, 2008), is useful for generating knowledge needed. To explore the experiences of Japanese emerging adults with SB, descriptive qualitative research was conducted. The function of descriptive qualitative research is to identify "what exists in the

social world and the way it manifests itself” (Ritchie, 2003) and describe it in detail from the study participants’ perspective. By using this research method, the researcher will be able to explore what Japanese emerging adults with SB experience during transition and consider what the next steps are for facilitating their healthy transition to adulthood in the Japanese socio-cultural and healthcare environment.

### **Conceptual Definitions**

Concepts addressed in this study are emerging adulthood, transition to adulthood, and self-management. As this study explores the experiences of Japanese emerging adults with SB, the concepts need to be defined in the context of Japanese socio-cultural context.

#### **Emerging Adulthood**

Emerging adulthood is a concept proposed by Arnett (2000). The concept was proposed to distinguish a developmental stage that ranges from the late teens through the twenties, typically ages 18-29 years, from adolescence or young adulthood (Arnett, 2000, 2015). This developmental stage has resulted from the social and demographic changes that occurred in 1960s and 1970s mainly in developed countries, such as increase in postsecondary education enrollment and later entry to marriage and parenthood (Arnett, 2015). According to Arnett (2015), there are five features that are distinctive to emerging adulthood: “identity explorations, instability, self-focus, feeling in-between, and possibilities/optimism” (p. 9). During this life stage, young people start to accomplish more independence than in adolescence and explore different possible choices in adult life (Arnett, 2000, 2015).

Emerging adulthood likely exists in Japan, too. Several demographic trends support this assertion. First, ages at the first marriage are constantly rising for both men and women. The

average ages of the first marriage are 31.1 years for men and 29.4 for women in 2014 whereas they were 26.1 years and 22.9 years respectively in 1947 (Minister of Health, Labour and Welfare, 2016). Age of entry into parenthood is also rising. The average age at first birth for Japanese women was 30.6 years in 2014 whereas it was 24.4 in 1950 and 25.6 in 1970 (Ministry of Health, Labour and Welfare, 2016). Second, college education is increasingly common among Japanese youth. In 2015, 79.8% of Japanese 18-year-old youth went on to higher education institutions, including two-year and four-year colleges, technical colleges, and higher professional schools (Ministry of Education, Culture, Sports, Science and Technology, 2015). The percentage of youth who went on to two-year or four-year colleges was 56.5%; the proportion has become more than five times higher than the proportion in 1955, which was 10.1%.

In Japan, therefore, the social and demographic changes appear to have occurred, thus allowing young people to spend their late teen and twenties for the purposes other than taking on adult roles. It is likely that Japanese youth in the above-mentioned age range experience an age period that is characterized as more independent than adolescence and more instable and uncertain than adulthood.

### **Transition to Adulthood**

In the context of the life-span development and health of individuals with SB, the concept of transition to adulthood consists of two components: transition to adult life and transition from child-centered to adult-centered health care. These two components are elaborated below.

**Transition to adult life.** According to Arnett (2003), an adult is a person who is able

to “stand alone as a self-sufficient person (p. 63).” As mentioned above, Arnett (2000, 2015) implies the presence of developmental milestones typically achieved in adulthood, such as marriage, entry into parenthood, and to have a regular job. Although these milestones could be universal across cultures, there appear to be commonalities and differences between Japanese and North American youth in the developmental tasks that are considered to be important to achieve during transition to adulthood.

Some Japanese researchers conducted studies to compare what independence as an adult means in Japan with what it means in the United States (Suzuki, 1999; Yamada & Miyashita, 2007). One study showed that both Japanese and the U.S. youth considered the following aspects as important for independence as an adult: ability to take care of oneself, ability to make decisions by oneself, ability to talk to parents on an equal basis, and to be financially independent (Suzuki, 1999). On the other hand, youth in the United States attached more importance to leaving parental home than Japanese youth while Japanese youth considered marriage as slightly more important than U.S. youth (Suzuki, 1999).

When defining transition to adult life in this study, the socio-cultural environment surrounding Japanese emerging adults with SB should be taken into consideration since it may be different from Western societies in some respects. For example, a Japanese psychiatrist Takeo Doi (2001) analyzed the characteristics of Japanese people’s interpersonal relationships using a unique Japanese word *amae*, which bridges the concepts of “dependence” and “attachment” (Doi, 1992). Doi (2001) maintained that dependence on parents tends to be accepted in Japanese society due to societal acceptance of *amae*. Such cultural context may considerably affect transition processes for emerging adults with SB.

**Transition from child-centered to adult-centered health care.** Transition from child-centered to adult-centered health care is a dynamic, life-long process that occurs for the purpose of fulfilling the needs of the individuals with special health care needs in a way appropriate for their developmental stages (American Academy of Pediatrics et al., 2002). In some cases transfer of care from a child-centered to an adult-centered health care provider occurs while in other cases children continue to receive care from the same health care provider with changing care needs that reflect their developmental stages (American Academy of Pediatrics et al., 2002). The goal of the health care transition is “to maximize lifelong functioning and potential through the provision of high-quality, developmentally appropriate health care services that continue uninterrupted as the individual moves from adolescence to adulthood” (American Academy of Pediatrics et al., 2002, p. 1304).

Binks, Barden, Burke, and Young (2007) conducted a literature review on transition to adult-centered health care for individuals with SB or cerebral palsy. They identified five key elements for positive transition: preparation, flexible timing, care coordination, transition clinic visits, and interested adult-centered health care providers. According to Binks et al. (2007), little is known about actual process of transition although studies have revealed unpreparedness of the patients and caregivers in adult health care settings and limited opportunity to receive guidance from health care providers.

### **Self-Management**

Self-management is a complex phenomenon central to the experiences of individuals with chronic conditions and their families (Ryan & Sawin, 2009). For emerging adults with SB, self-management is deeply connected with transition to adulthood and may be understood more

fully by taking families into account due to dynamic change in roles and responsibilities of the emerging adult and his/her family members during transition. The Individual and Family Self-Management Theory (IFSMT) defines self-management as:

A process by which individuals and families use knowledge and beliefs, self-regulation skills and abilities, and social facilitation to achieve health-related outcomes. Self-management takes place in the context of risk and protective factors specific to the condition, physical and social environment, and individual and family. Proximal outcomes are self-management behaviors and cost of health care services; distal outcomes are health status, quality of life and cost of health. (Ryan, 2009; Ryan & Sawin, 2009, 2014; UWM Self-Management Science Center, 2021; UWM Self-Management Science Center Working Group, 2011 retrieved from [www.uwm.edu/nursing/research/self-managment](http://www.uwm.edu/nursing/research/self-managment)).

In light of these condition-related and theoretical contexts, self-management behaviors in emerging adults with SB are behaviors that facilitate management of existing symptoms due to this congenital condition and prevention or alleviation of secondary conditions, including but not limited to neurogenic bowel and bladder, skin conditions, musculoskeletal conditions, orthopedic conditions, and seizures.

The influence of social environment and familial context on individual and family self-management behaviors proposed in the IFSMT is particularly important when studying experience of Japanese emerging adults with SB during transition. Family factors within Japanese socio-cultural context may strongly affect self-management of the emerging adults with SB. For example, according to Hori, Narama, and Yamauchi (2002), levels of independence in

some self-management behaviors tend to be influenced by parenting attitudes. Also, it appears difficult even for Japanese mothers of middle-school children to entrust their children to do enema and skin care by themselves (Hori et al., 2002). This difficulty for the parents to let go of control over their children's self-management may be associated with Japanese socio-cultural context that is relatively tolerant of dependence on others. The self-management of Japanese emerging adults with SB, therefore, needs to be understood in conjunction with the socio-cultural and familial contexts.

## **Overview of the Study**

### **Purpose of the Study and Research Questions**

The purpose of this study was to describe the experiences of transition to adulthood for Japanese emerging adults with SB. The research question for the study was “What are the experiences of transition to adulthood for Japanese emerging adults with SB?”

### **Theoretical Frameworks**

Three theoretical approaches influenced the development of this study: the theory of emerging adulthood, the Ecological Model of Secondary Conditions and Adaptation, and the Individual and Family Self-Management Theory. Emerging adulthood is defined as a developmental stage that ranges from the late teens through the twenties, typically ages 18-29 years, when youths start to accomplish more independence and explore different possible career choices in adult life (Arnett, 2000, 2015). Although commonalities are likely to exist between emerging adults and adolescents with regard to transition experiences, there are several significant developmental milestones regardless of whether he/she has illness or disability that distinguish emerging adulthood from younger ages in Japan, such as eligibility for driver's

license and college entry as well as the voting age, which was lowered from 20 to 18 in 2016. Therefore, by focusing on emerging adulthood, the researcher will be able to better understand unique experiences of the individuals in the developmental stage in which youth move into adulthood.

The Ecological Model of Secondary Conditions and Adaptation (Sawin et al., 2003; Sawin & Thompson, 2009) was also a framework informing this study. The ecological model proposes that adaptation during transition in adolescents and young adults with SB is associated with both risk factors and protective factors (Sawin et al., 2003). The risk factors include: (a) demographic risk factors, e.g., age, gender, and socioeconomic status, (b) condition-specific risk factors, e.g., severity of illness, level of lesion, and presence of hydrocephalus, and (c) neuropsychological impairments, e.g., executive functioning and working memory (Sawin et al., 2003; Sawin & Thompson, 2009). The protective factors include: (a) adolescent resilience, e.g., decision-making maturity, household responsibility, beliefs, and communication efficacy, (b) family resourcefulness, e.g., family activities, family cohesion, and satisfaction, and (c) health care adequacy (Sawin et al., 2003; Sawin & Thompson, 2009). The adaptation outcomes in this model refer to physical health, mental health, social competence, academic achievement, and quality of life (Sawin et al., 2003; Sawin & Thompson, 2009).

The third theoretical framework informing this study was the Individual and Family Self-Management Theory (Ryan, 2009; Ryan & Sawin, 2009, 2014; UWM Self-Management Science Center, 2021). In this theory, self-management is proposed to have three dimensions: context, process, and outcomes. The context dimension refers to risk and protective factors, which can be categorized into condition-specific factors, physical and social environments, and

individual and family factors (Ryan & Sawin, 2009; UWM Self-Management Science Center, 2021). The process dimension refers to the process of behavior change, including the impact of knowledge and beliefs, self-regulation, and social facilitation (Ryan & Sawin, 2009; UWM Self-Management Science Center, 2021). The outcomes dimension refers to both proximal and distal results of the self-management process. Proximal outcomes refer to self-management behaviors and cost of health care services, and distal outcomes refer to health status, quality of life, and cost of health (Ryan & Sawin, 2009; UWM Self-Management Science Center, 2021).

These frameworks influenced the development of a flexible interview guide. The goal of the interview guide was to facilitate the participants' reflection on potentially relevant areas in their experiences during transition to adulthood.

### **Sample**

The participants of the study included eight Japanese emerging adults with SB aged 20 to 29 years who live in Japan. The participants were recruited using purposeful sampling method to assure relevance of the data, homogeneity of the sample in terms of the types and levels of the condition and disability, and the variation in the participants' experiences. The recruitment was conducted through the Spina Bifida Association of Japan.

### **Design**

This study used a qualitative approach to describe the experiences of Japanese emerging adults with SB during transition to adulthood to understand the emerging adults' experiences and the meanings attached to them in detail. This approach was chosen because, as mentioned above, transition to adulthood for emerging adults with SB is a phenomenon that is newly developing as medical management of the condition advances. The philosophical perspective

guiding knowledge development in this study was social constructivism. In the light of social constructivism, the researcher is dependent on the participants' views when obtaining knowledge (Creswell, 2006); the purpose of knowledge development is to understand subjective experiences emerging in the participants' minds and the meanings they construct (Lincoln & Guba, 1985).

The qualitative descriptive method suited the purpose of the study because (a) the participants' experiences during transition were complex and therefore they needed to be understood as a whole and (b) it was important for the experiences to be understood from the perspectives of the emerging adults. For the same reasons, an in-depth interview was a suitable data collection method for listening to and understanding the stories about the participants' experiences during transition to adulthood. By qualitatively analyzing the data from in-depth interviews with the Japanese emerging adults, themes describing their experiences were identified and variations in their experiences were explored.

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## CHAPTER TWO

### Synthesis of Literature

The proposed population of interest for this dissertation is Japanese youth with spina bifida (SB) who are transitioning to adulthood. This chapter consists of two literature review manuscripts: (a) a synthesis of the international literature on transition for adolescents and emerging adults (AEA) with SB into adulthood written in English and (b) a synthesis of the literature on AEA with SB published in Japanese journals.

It is important to review both studies written in English and studies published in Japanese journals for the following two reasons. First, the characteristics of the experiences of Japanese adolescents and emerging adults with SB during transition could be better understood by comparing their experiences with that of adolescents and emerging adults in other countries. The experiences of these adolescents and emerging adults may be influenced by various socio-cultural contexts, such as culturally desirable family relationship, education systems, healthcare systems, and welfare programs for individuals with chronic illness and disability. Therefore, it is important to identify what is already known about the differences and commonalities between the experiences of the adolescents and emerging adults in Japan and in other areas of the world, such as Europe and North America. Second, comparison of the current status of studies published in Japanese journals with studies written in English will facilitate the identification of gaps in the literature and areas where further research is needed concerning Japanese AEA with SB.

In this chapter, the synthesis of the literature on AEA with SB written in English is presented first. Second, the synthesis of the literature on adolescents and emerging adults with

SB published in Japanese journals is reported. Comparison between the findings of the two syntheses is presented at the end of the chapter.

**Manuscript #A. The Transition Experiences of Adolescents and Emerging Adults with Spina Bifida: A Synthesis of Literature Written in English**

**Abstract**

**Background:** Transition to adulthood has become one of the significant issues for adolescents and emerging adults (AEA) with spina bifida (SB) as their life expectancy has been dramatically improved. To explore the state of the science on experiences of transition for adolescents and emerging adults with SB into adulthood, a synthesis of the literature written in English was conducted.

**Methods:** MEDLINE and CINAHL databases were searched using the following key words: “spina bifida” or “myelomeningocele” and “transition.” Research articles published during the years 2004 and 2018 in English were included.

**Results:** The final sample consisted of 45 research articles. Four themes were identified: (a) transition experience of AEA with SB, (b) transition outcomes, (c) services currently offered and service needs for AEA with SB, and (d) transition to adult health care.

**Conclusions:** Knowledge gaps and implications for future research and for nursing practice were discussed. Research with more powerful design, as well as additional qualitative studies that assist health care providers to understand the experience of the AEA, are needed to improve clinical practice for the population.

*Keywords:* spina bifida, adolescents, emerging adults, transition

Spina bifida (SB) is a congenital condition characterized by a lack of neural tube closure during fetal development. The survival rate among infants born with SB has been improving dramatically and continuously worldwide (Wong & Paulozzi, 2001). Consequently, transition to adulthood has emerged as one of the most significant issues for individuals with SB and their families. In most cases individuals with SB have normal cognitive abilities (National Institute of Neurological Disorders and Stroke, 2014) ; however, physical impairments and learning disabilities may increase the complexity of their condition. As a result, adolescents and emerging adults (AEA) with the condition often experience difficulty in accomplishing desirable developmental outcomes due to their impairments as well as environmental barriers. Investigators have just begun to describe the transition experiences and outcomes in AEA with SB.

Two literature reviews (Binks et al., 2007; Snow-Lisy et al., 2015) have been conducted on transition to adult health care for AEA with SB or cerebral palsy. Binks et al.'s review (2007) identified barriers related to transition for AEA with SB or cerebral palsy and key factors for their positive transition. Newer evidence on these factors needs to be reviewed. More recently, Snow-Lisy et al. (2015) synthesized research literature on urological management of SB. Some psychological and social issues individuals with SB experience during transition, such as depression, mobility issues, problems of school system, and low percentage of independent living status, were identified; however, the findings were focused on medical management of SB and the role of pediatric urologists (Snow-Lisy et al., 2015), thus limiting the scope of the review.

In addition, programs with the purpose of facilitating the transition process for

adolescents and young adults with chronic conditions have been developed in various regions of the world. Although many of those programs still appear to be under development, the literature on such programs may also provide useful information for improving transition services for the population.

### **Purpose**

The primary purpose of this chapter is to explore the state of the science on experiences of transition for AEA with SB into adulthood through a synthesis of the literature written in English and published in international journals. Critiques of the design and samples of the studies are provided. In addition, implications for future research and for nursing practice are discussed.

### **Methods**

#### **Literature Search Methods**

A synthesis of research literature on transition for AEA with SB into adulthood was conducted. The search strategy involved database search for research studies using MEDLINE and CINAHL. The key words used for the database search included “spina bifida” or “myelomeningocele” and “transition.” Manual searching of reference lists of articles found was also conducted to identify additional relevant studies for review.

Inclusion criteria for this literature review were research studies: (a) with AEA or parent as participants, (b) written in English, (c) published during the years between 2004 and 2018 and (d) that focused on the experiences of AEA during transition into adulthood, including but not limited to self-management, social participation, mental health, psychosocial issues, quality of life, transition outcomes, and transition programs, and (e) that used a sample that consisted of

multiple diagnostic groups if AEA with SB were specifically included. Studies on urological problems in AEA that addressed not only physical conditions but also psychosocial issues were included. Articles on experiences with adult health care or transition from pediatric to adult health care were included in the sample. Exclusion criteria included (a) articles published before 2004, (b) articles that were not peer-reviewed research reports, (c) dissertations, (d) single case studies, (e) studies that addressed only epidemiological aspect of SB, and (f) studies that addressed only specific physical or neurocognitive functions.

In this literature review, the topic of interest was studies on transition for AEA with SB written in English. An adolescent was defined as a young person aged between 10 and 19 years (World Health Organization, 2014) who is in the developmental phase characterized by dramatic changes both in the self and in their relationships with their contexts (Lerner & Galambos, 1998). The definition of emerging adult was derived from Arnett's (Arnett, 2000, 2015) theory of emerging adulthood. An emerging adult was defined as a person aged between 18 and 29 years who is in the developmental period of exploration for different future directions after leaving the period of dependency (Arnett, 2015).

### **Selection and Analysis of Studies**

One hundred and forty-six articles were identified through the database search with MEDLINE and CINAHL. Twenty-six duplicate articles were identified and excluded. After the remaining 120 articles were reviewed to check for consistency with the inclusion criteria, 80 articles were excluded. Five articles were identified through hand searching of reference lists and added to the sample. As a result, 45 research articles from Europe and North America were included in the final sample (see Figure 2.1).

In conducting the review of the full texts of the sample, the following information was extracted and summarized in an Evidence Table: study purpose, design, framework, sample, measures, analysis, findings, strengths, and limitations (See Table 2.1). Based on the review, common themes addressed in the studies were identified. The guideline for assessing level of evidence developed by Melnyk and Fineout-Overholt (2011) was used to evaluate the quality of the studies.

### **Findings and Discussion**

The review of the literature identified four themes: overall transition experience of AEA with SB, challenges to achieving transition outcomes, barriers to transition to adult health care, and services needed and available services for AEA with SB. Summaries of the four themes are provided in the following sections.

#### **Overall Transition Experiences of Adolescents and Emerging Adults**

Nine studies addressed experience of AEA with SB during transition (Buran et al., 2004; Heller et al., 2016; Kinavey, 2006, 2007; Levy et al., 2014; Lindsay et al., 2017; Ridosh et al., 2011; Sawin et al., 2009; Stephany et al., 2015). Subtopics addressed in these articles included self-management (Buran et al., 2004; Ridosh et al., 2011; Sawin et al., 2009), independence (Ridosh et al., 2011; Sawin et al., 2009), stigma (Lindsay et al., 2017; Ridosh et al., 2011; Sawin et al., 2009), inner strength (Ridosh et al., 2011), advocacy (Ridosh et al., 2011; Sawin et al., 2009), professional and social support for school-work transition (Lindsay et al., 2017), romantic and sexual relationships (Heller et al., 2016), exploration of identity (Kinavey, 2006, 2007), and preparing for life as an adult with SB (Strömfors et al., 2017). Also, the recurring themes identified in focus group interviews with Scottish families who have young patients with SB

included issues concerning communications, respect, choice, and control during transition (Levy et al., 2014). Those nine studies mainly used qualitative research methods to describe or explain the experiences of the AEA.

The experiences of self-management were described in relation to various influential factors, including knowledge, skills, aspirations, and parenting styles (Sawin et al., 2009). Self-management behaviors have been reported as low in adolescents with SB (Buran et al., 2004). Two studies delineated how the AEA learned to practice self-management skills and activities of daily living, thus pursuing independence (Ridosh et al., 2011; Sawin et al., 2009). Those two studies also revealed that limited social interaction, discrimination, and stigma were involved in the AEA's self-management experiences, and inner strength and self-advocacy as well as peer and adult advocacy could be protective factors to increase resilience during transition (Ridosh et al., 2011; Sawin et al., 2009).

Identity exploration and formation for late-stage adolescents with SB was investigated in two studies (Kinavey, 2006, 2007). Kinavey (2006) identified three primary ways in which the study participants assigned meaning to their disability: (a) identity as overcoming disability; (b) identity objectifying disability; and (c) identity as integrating disability. She also found that the participants described in common the theme of "otherness" (Kinavey, 2007, p. 154) that they perceived with regard to visible physical differences and use of wheelchairs. This sense of otherness may exacerbate social isolation, thereby preventing adolescents from receiving social support both at home and in a community. In addition, Heller et al. (2016) explored the experiences related to romantic and sexual relationship in AEA with SB, revealing their challenges with being involved in romantic relationship and discussing sexuality and the

condition with the partner.

### **Challenges to Achieving Transition Outcomes**

Eleven studies examined possible outcomes of transition for AEA with SB to adulthood (Barf et al., 2007; Bellin et al., 2011; Buran et al., 2004; Cox et al., 2011; Dicianno et al., 2009; Galambos et al., 2008; Heffelfinger et al., 2008; Johnson et al., 2007; Liptak et al., 2010; Woodward et al., 2012; Young et al., 2006). The outcomes examined included functional independence (Buran et al., 2004; Heffelfinger et al., 2008), self-management (Bellin et al., 2011; Buran et al., 2004), developmental competence (Buran et al., 2004), psychosocial maturity (Galambos et al., 2008), independent living (Bellin et al., 2011; Young et al., 2006), social participation including attending schools and employment (Bellin et al., 2011; Dicianno et al., 2009; Johnson et al., 2007; Liptak et al., 2010), psychological distress (Dicianno et al., 2009), quality of life (QOL) (Bellin et al., 2011; Cox et al., 2011; Dicianno et al., 2009), and life satisfaction (Barf et al., 2007).

These studies examined current conditions regarding the transition outcomes and/or factors associated with them. The adolescents with SB in these studies did not demonstrate adequate engagement in decision-making, responsibility, or activities with peers and needed moderate levels of assistance with self-management tasks (Buran et al., 2004). Likewise, in the United States, among 87 youth ages 11 to 22 years with chronic conditions including SB, 85% were living with parents or siblings (Woodward et al., 2012). The factors associated with functional independence included age, level of lesion, executive functioning, skills to initiate self-management routines independently, and adolescent activities, such as chores and decision-making (Heffelfinger et al., 2008). Psychosocial maturity was associated with use of task-

focused coping, lower level of depression, and perception of fathers' fostering of autonomy (Galambos et al., 2008). Gender and self-management were associated with independent living (Bellin et al., 2011). Level of lesion, gender, ethnic groups, speaking a language other than English were associated with social participation (Bellin et al., 2011; Liptak et al., 2010).

Neither urological nor neurosurgical outcomes were associated with QOL (Cox et al., 2011). However, mobility status was significantly associated with lower QOL while not associated with self-management and psychological distress (Dicianno et al., 2009). Development of a tool to assess QOL specifically for adolescents and young adults with disability may facilitate generation of evidence concerning the transition outcomes. A questionnaire to measure QOL for individuals with disability transitioning to adulthood has been developed and validated with a sample of adolescents and young adults including those with spina bifida (Deroche et al., 2015). As a result of an exploratory factor analysis, the five factors characterizing QOL for these individuals included emotional health, physical health, independence, activity limitation, and community participation (Deroche et al., 2015).

Life satisfaction was also examined as a transition outcome. The proportion of the AEA with SB who reported dissatisfaction with life as a whole in Barf et al.'s study (2007) was 24%. No significant associations were found between overall life satisfaction and demographic and condition-related variables examined (Barf et al., 2007). However, AEA with SB and hydrocephalus were less satisfied with their self-care ability and relationship with partner compared to those without hydrocephalus and reference group (Barf et al., 2007).

In summary, a variety of transition outcomes were proposed in the literature. The existing international literature published in English indicates that delayed transition patterns are

observed among AEA with SB. Various individual- and family-level factors are associated with these transition outcomes.

### **Barriers to Transition to Adult Health Care**

A total of 20 studies addressed transition to adult health care for adolescents and young adults with SB. Two of these were experimental or quasi-experimental studies that involved implementation of transition preparation programs (Betz et al., 2010; Sawin et al., 2015). In addition, one article explored reasons for participation in a transition preparation program (Smith et al., 2015). There were two other articles that descriptively reported implementation of specific transition programs for AEA with SB (Berens & Peacock, 2015; Kaufmann Rauen et al., 2013; Ruck & Dahan-Oliel, 2010). Five studies specifically addressed urologic problems and their management during transitional period (Shalaby et al., 2015; Summers et al., 2014; van der Toorn et al., 2013). Seven studies explored readiness for and experiences of transition to adult health care and identified barriers, facilitators, and solutions of health care transition (Berry et al., 2013; Grimsby et al., 2016; van Staa et al., 2011; Young et al., 2009) or described the process of health care transition with regard to medical record management (Østerlund et al., 2005). Two studies addressed health outcomes in AEA during the period of transition to adult health care.

A randomized controlled trial was conducted to test the effectiveness of a cognitive-behavioral program, Transition Preparation Training, for facilitating transition in adolescents with SB (Betz et al., 2010). The program emphasized the development of “an adolescent-centered transition plan” (Betz et al., 2010, p. 598) for assisting adolescents to identify and obtain necessary services based on their future goals. There were no significant differences

between groups of any treatment effect, i.e., subjective well-being, role mastery, or self-care practice (Betz et al., 2010). Betz et al. argued that this lack of significant results could be attributed to insufficient intervention dose, adolescents' lack of adherence, and lack of empirically sound instruments to measure the outcomes. The reasons that the adolescents participated in the intervention study were investigated (Smith et al., 2015). The adolescents' desire to learn more about their condition and altruistic values were commonly reported (Smith et al., 2015).

The second study, an early evaluation of a new transition program, the Spina Bifida Transition Project, documented that the program, which was developed based on best practice and involved many practical components, was positively perceived by the young adults and their parents (Sawin et al., 2015). The program was guided by the Ecological Model of Secondary Conditions and Adaptation in Spina bifida (Sawin et al., 2003) and the Transition Care Model (Kaufmann Rauen et al., 2013) which described elements of building a bridge between pediatric to adult health care. The program was developed collaboratively by both pediatric health care and adult health care professionals and designed to facilitate preparation for smoother transition to adult health care, which included AEA and families' tour to adult clinic visit and preparation of an individual clinical summary (Kaufmann Rauen et al., 2013). The emerging adults and parents alike indicated that a major unanticipated experience was that the emerging adults was "treated like an adult", which despite the best intentions of pediatric providers, did not occur in the pediatric setting, even if the patient was an adult. In a study that evaluated a transitional care using descriptive design, a multidisciplinary program that involved regular monitoring and evaluation of transition processes was rated overall satisfactory by AEA with SB in terms of

teen-centeredness (Ruck & Dahan-Oliel, 2010). Patient population of an academic primary care clinic for AEA with chronic conditions was described in another study (Berens & Peacock, 2015). AEA with SB accounted for 16% of the total patient population of the clinic, and specialties most commonly utilized among AEA with SB included urology (59.6%), neurosurgery (28.8%) and physical/occupational therapy (28.8%) (Berens & Peacock, 2015). A review of these evaluation reports revealed that the components of the transition programs were often influenced by Binks et al.'s (2007) systematic review.

Although the results of these three studies cannot be simply compared due to the difference in research methods, ages, and program structures, in Sawin et al.'s (2015) and Ruck and Dahan-Oliel's (2010) studies the AEA and their families appraised the programs overall satisfactory while no significant differences in outcomes were found in Betz et al.'s (2010) randomized controlled trial. One possible reason for this difference may be, as Betz et al. (2010) pointed out, the characteristics of the samples. In Betz et al.'s study, the sample mainly consisted of younger Latino adolescents with low socioeconomic status preparing for transition while more than 90% of the participants were Caucasian emerging adults in Sawin et al.'s (2015) study. For adolescents and their families with low socioeconomic status, it may have been difficult to adhere to the program (Betz et al., 2010). Unmet needs of such adolescents and their families need to be explored to improve transition preparation programs for AEA whose ethnic and socioeconomic backgrounds are diverse. The studies also examined different outcomes. Sawin et al (2015) were interested in older adolescents and emerging adults and their parent's satisfaction with transfer to adult healthcare whereas Betz et al. attempted to increase self-care and independence in a younger cohort still in a pediatric health care setting.

In addition, research has been accumulating concerning urologic problems in individuals with SB and other complex genitourinary conditions during transition to adult health care. Urologic problems these individuals commonly experience included urinary incontinence, recurrent urinary tract infections, and catheterization challenges (Summers et al., 2014). Urology clinics providing transitional care have been introduced in the United States (Shalaby et al., 2015). At an adolescent transition urology clinic, adolescents who require long-term urologic management are seen by both pediatric and adult urologists as well as a urology nurse (Shalaby et al., 2015). The majority of these adolescents reported that the services provided at the clinic were beneficial to prepare them for self-management in transition (Shalaby et al., 2015).

The experiences of transition to adult health care for AEA with SB were explored in two quantitative (Grimsby et al., 2016; Stephany et al., 2015), five qualitative studies (Berry et al., 2013; Hartman et al., 2018; Lindsay et al., 2016; van Staa et al., 2011; Young et al., 2009), and one quality improvement study with both quantitative and qualitative data (Squiers et al., 2017) in which both barriers and facilitators for transition processes were identified. Barriers to transition to adult health care identified in these studies included parents' reluctance and lack of awareness of need to leave pediatric care, uncertainty of the transition process, lack of information, lack of access to health care, cultural gaps between pediatric and adult services, lack of support from primary care providers, and insurance coverage issues (Berry et al., 2013; Grimsby et al., 2016; van Staa et al., 2011; Young et al., 2009). In a study on AEA's and family's readiness regarding transition with a sample of 15 AEA ages 14 to 31 years, 40% of AEA reported that they never wanted to transition and their interest in transition was weaker than

their parents (Stephany et al., 2015) Facilitators included youth's higher cognitive ability, youth's strong interest in independence, parenting style that fosters independence, and success in negotiation with familiar adult health providers (Berry et al., 2013). Those barriers and facilitators were congruent with the findings of the systematic review (Binks et al., 2007).

Also, health outcomes, such as health service use and the development of secondary conditions during the period of transition to adult health care were examined in two studies (Mahmood et al., 2011; Mann et al., 2015). The rates of emergency room visits and inpatient hospitalization for AEA with SB were significantly higher than for the comparison group (Mann et al., 2015). Moreover, young adults with SB aged between 20 and 24 years were at a higher risk of emergency room visits compared to adolescents aged between 15 and 19 years (Mann et al., 2015). Lower level of self-management behaviors and higher number of ulcers were significant predictors of hospitalizations (Mahmood et al., 2011). Multivariate analyses revealed that history of shunting, lower education level, self-management behaviors, and employment status were significantly associated with urinary tract infections (Mahmood et al., 2011). Also, higher education level and motor level but not satisfaction with care received were significantly associated with pressure ulcers (Mahmood et al., 2011).

### **Services Currently Offered and Service Needs for Adolescents and Emerging Adults with Spina Bifida**

Three studies addressed services for AEA with SB during transition (Bjornson et al., 2011; Darrah et al., 2010; Woodward et al., 2012). In one study parents of AEA with special health care needs including SB reported services most needed were dental care (95%), routine preventive care (95%), prescription medications (93%), and specialty care (78%) whereas they

reported that their AEA's highest unmet needs were physical, occupational, and speech therapy (26%) (Woodward et al., 2012). Emerging adults who had received physical and/or occupational therapy in adolescence demonstrated higher levels of participation in postsecondary education whereas receiving the therapy was not associated with paid employment (Bjornson et al., 2011). On the other hand, community services including educational, employment, transportation, and assured income programs designed to facilitate independence were paradoxically found to restrict independence and options for social participation among emerging adults with physical disabilities (Darrah et al., 2010). Interestingly, health care was identified as important and generally the need was met for emerging adults participating in one transition program. However, participants identified services that facilitated housing and employment as unmet needs (Sawin et al., 2015).

## **Quality Assessment**

### **Study Design and Sample**

The sample of 45 studies included 25 quantitative studies, 16 qualitative studies, two studies using both quantitative and qualitative methods, and two literature reviews. The quantitative studies consisted of one experimental study, two prospective cohort studies, one case-control study, and 21 descriptive, correlational studies. The experimental study employed randomized controlled trial design to test the effectiveness of a cognitive-behavioral program in facilitating transition (Betz et al., 2010). Using the rating system of evidence developed by Melnyk and Fineout-Overholt (2011), the sample for this review included one experimental study at level II, two prospective cohort studies at level IV, one case-control study at level IV, and two literature reviews at level V while the remaining studies were rated at level VI (evidence

from a single qualitative or descriptive study).

The methods used in the qualitative studies included narrative analysis (Ridosh et al., 2011), grounded theory (Østerlund et al., 2005), phenomenological ethnography (Kinavey, 2006, 2007), content analysis approach (Darrah et al., 2010; Sawin et al., 2009; Smith et al., 2015), and thematic analysis (Sawin et al., 2015; van Staa et al., 2011). The investigators of the other three qualitative studies did not explicitly identify specific research methodology.

Overall, strategies to enhance rigor were presented in detail in the qualitative research reports reviewed, while small sample size (Berry et al., 2013; Kinavey, 2006, 2007), lack of diversity of the sample (Ridosh et al., 2011; Sawin et al., 2009), and possible underrepresentation of a specific subgroup (Berry et al., 2013; Sawin et al., 2015; Østerlund et al., 2005) may have compromised transferability of the study findings (Lincoln & Guba, 1985). Several methodological issues limited the quantitative studies reviewed. First, although the sample for the current review included a randomized controlled trial (Betz et al., 2010), most studies employed correlational designs. Therefore, those studies are limited with regard to their ability to demonstrate causal relationships. Second, in the majority of the studies participants were recruited at a single research site, thus limiting the generalizability of the findings. Finally, low response rates may have decreased the sample representativeness in some studies (Cox et al., 2011; Levy et al., 2014; Woodward et al., 2012).

In addition, there were some studies that used samples consisting of multiple diagnostic groups (Bjornson et al., 2011; Darrah et al., 2010; Galambos et al., 2008; van Staa et al., 2011; Woodward et al., 2012; Young et al., 2006; Young et al., 2009). Although to be selected for the current review the samples included AEA with SB and some studies analyzed each diagnostic

group separately, in most cases results were presented for the total sample. Thus the generalizability of those studies to individuals with SB may be limited.

Finally, it should be noted here that the experiences and psychosocial issues of AEA with SB vary considerably depending the type of the condition and level of lesion. Even when the sample consisted of only individuals with SB, the sample may not have been a homogeneous sample. Therefore, the findings of the current review should be interpreted with caution.

### **Knowledge Gaps and Implication for Future Research**

The current analysis of manuscripts on experience of AEA with SB published in English highlighted gaps and future research directions. First, empirical knowledge concerning the effectiveness of transition intervention programs for AEA with SB has not been sufficiently developed. Also, it appears that the existing experimental study was not explicitly guided by a theory (Betz et al., 2010) while the other program evaluation was guided by the Transition Care Model and the Ecological Model of Secondary Conditions and Adaptation in Spina Bifida (Kaufmann Rauen et al., 2013). More experimental studies that assess the effectiveness of the programs based on theoretical framework will be needed.

Second, the sample for this review included only two studies with a prospective design (Bjornson et al., 2011; Liptak et al., 2010); the remaining quantitative, non-experimental studies all used a retrospective design. Although the use of a proxy measure (e.g., wheelchair use as a proxy variable of walking ability due to secondary analysis) may become a threat to validity in this secondary analysis, the powerful design enabled the demonstration of the association between rehabilitation services and positive transition outcomes (Bjornson et al., 2011). Future research will need to employ more powerful designs to generate evidence useful for clinical

practice.

Third, research literature that addressed transition, its outcomes, and the factors related to the outcomes from the perspectives of AEA with SB is limited. There are two reasons for this gap. For one thing, some studies appeared to rely on parents' responses somewhat heavily in examining adolescents' and/or emerging adults' health and psychosocial status as well as service needs (Cox et al., 2011; Woodward et al., 2012). Those researchers indicated that they had to rely on parent proxy reports because the response rate of the youth was very low due to their impairment (Woodward et al., 2012). However, the agreement between youth' self-reports and parents' reports for psychosocial variables are often low (Buran et al., 2004) and creative strategies to collect self-report data from AEA should be developed for future research. For another, although various variables, such as functional independence, self-management, developmental competence, and psychosocial maturity, were used as transition outcomes in the studies reviewed, it is still unclear what the important outcomes are from the perspectives of AEA with SB. Research will be needed that examines what the AEA prioritize as outcomes of transition to adulthood.

### **Implication for Nursing Practice**

AEA with SB have aspirations for independence; however, they are often faced with challenges in pursuing it (Kinavey, 2006; Ridosh et al., 2011; Sawin et al., 2009). Generally, transition to adulthood is delayed in this population (Buran et al., 2004). The research literature has revealed several facilitators and barriers to transition. For example, age, gender, ethnic group, level of SB lesion, executive functioning, and adolescent activities, including chores and decision-making, were significantly associated with functional independence or social

participation as adaptation outcomes (Bellin et al., 2011; Heffelfinger et al., 2008; Liptak et al., 2010). Nurses caring for this population need to assess those factors as well as readiness for adulthood to identify the AEA strengths, resources, and areas that require support for facilitating transition.

Select studies on services for AEA with physical disabilities including SB supported the impact of that physical/occupational therapy during secondary education on positive transition outcomes whereas community services, such as employment training and transportation services paradoxically restricted their social participation (Darrah et al., 2010). Nurses need to pay attention to the interaction between the individuals' personal abilities and environmental contexts (Darrah et al., 2010). Also, services need to be more individualized and flexible (Darrah et al., 2010).

### **Limitations**

Use of "transition" as the primary keyword may have limited the sample of this review. Studies of early adolescents or specific issues, such as achievement of developmental tasks in adolescents may not have been identified in the database searches if the investigators did not use related terminology such as "autonomy." For example, Davis et al., (2006) detailed skills and behaviors achieved in those 12-18 years old with IQ of 80 or better as 2-5 years later than typically developing peers (TDP). It is not clear if this lag is developmentally appropriate for those with SB or due to lack of expectations and support. They found basic behaviors achieved by age 12 included dresses appropriately, making shopping choices, cleans room, knows phone number, and shops with assistance (Davis et al., 2006). Other behaviors such as saves money, cooks pre-packed meals, plans activities with peers and arranges own transportation were

generally achieved during adolescents but 2-5 years later than TDP (Davis et al., 2006).

Advanced behaviors achieved by TDP and generally not achieved by youth with SB by age 18 included does own laundry, cooks independently, manages bank account without assistance, manages allowance, makes own appointments. Participants did not generally enter adolescence with high levels of SB knowledge but developed them during adolescence (e.g., knowing signs of skin ulcer, bowel problems, shunt failure and urinary tract infection) (Davis et al., 2006). Future literature reviews could be more informative if related terms such as “autonomy” “self-efficacy” and “decision-making” are included in database searches.

### **Conclusions**

Forty-five research articles on transition from adolescence to adulthood for European and North American AEA with SB were reviewed. The results of the analysis indicated the following: (a) Studies on the overall experiences of transition to adulthood revealed that AEA with SB had aspirations for independence; however, they experienced various psychosocial challenges during transition and were not always successful in transition. (b) Various outcome variables were used in assessing transition outcomes and exploring factors associated with the outcomes. However, there is still much to be learned about the priority outcomes from the AEA perspective and the factors related to these outcomes. (c) Transition to adult health care has begun to be evaluated from multiple perspectives, but the only RCT of a transition program did not improve outcomes. Understanding what components of a health care transition program lead to successful transition outcomes is a major need. (d) Some rehabilitation services had a positive impact on AEA’s social participation whereas community services intended to facilitate independence had a negative impact on transition. Overall, experimental research and non-

experimental research with more powerful design, as well as additional qualitative studies that assist health care providers understand the lived experience of the EA, will be needed to improve clinical practice for the population.

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Figure 2.1.

Flow diagram of identification and selection of studies on experience of adolescents and emerging adults with spina bifida during transition written in English.

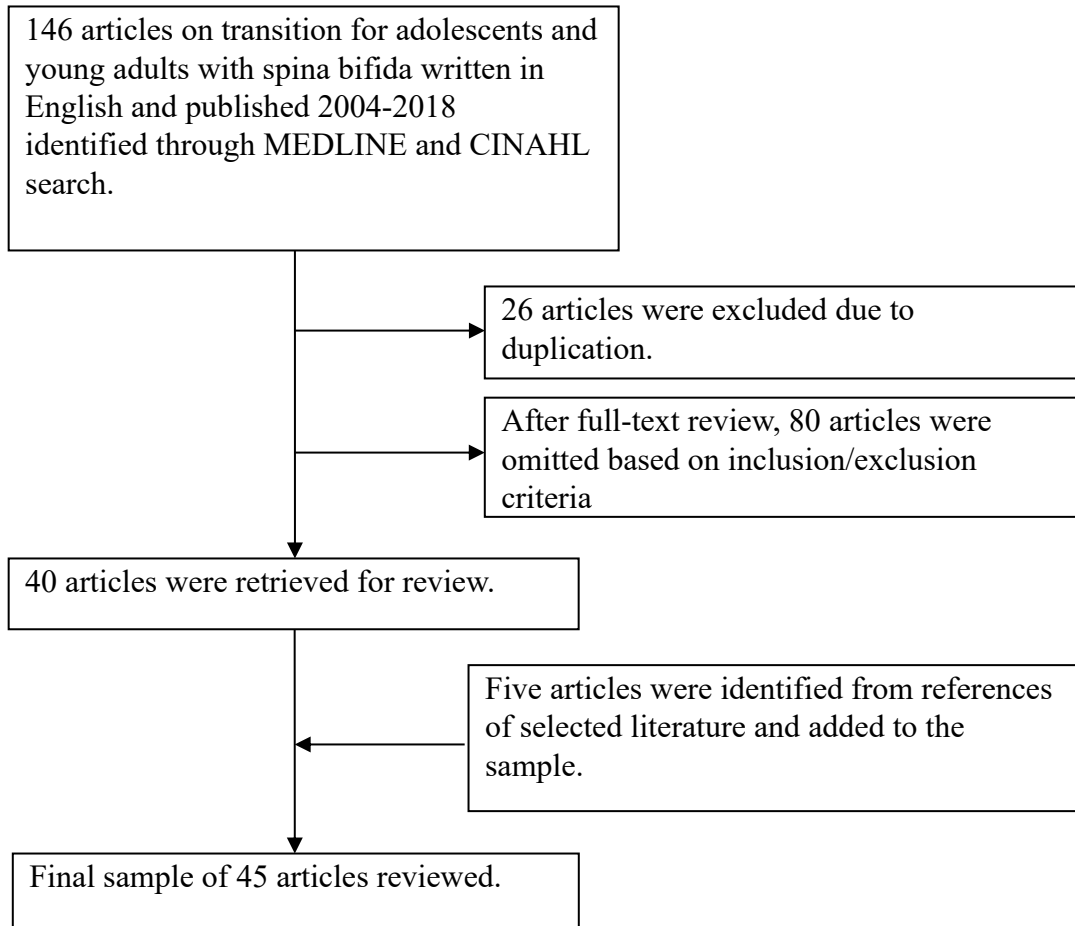


Table 2.1.

*Studies on Experience of Adolescents and Emerging Adults with Spina Bifida During Transition Written in English.*

Author(s)	Purpose/Aim	Design	Sample	Measures	Analysis Strategy	Relevant Results	Strengths/Limitations
Barf, Post, Verhoef, Jennekens-Schinkel, Goosken, & Prevo 2007 <b>Netherlands</b>	To investigate life satisfaction and its determinants in Dutch young adults with spina bifida.	Quantitative Cross-sectional  Level of Evidence: VI	179 young adults with spina bifida (ages 16-25yrs); 41% male; 67% hydrocephalus; 39% using wheel chair	Life Satisfaction Questionnaire, disease information (level of lesion, type of spina bifida, presence of hydrocephalus, ambulatory status, incontinence, and intelligence)	Descriptive statistics, logistic regression	21% were dissatisfied with their life. Young adults with hydrocephalus were more satisfied with their financial status and family life, but less satisfied with self-care and partnership relations than were those without hydrocephalus. <b>Logistic Regression:</b> Of the demographic and disease characteristics, higher level of lesion was associated with satisfaction with self-care ability (95%CI of odds ratio=1.65-9.53) and vocational/educational aspect of life satisfaction (95%CI of odds ratio=1.03-4.81). Having hydrocephalus was associated with satisfaction with financial situation (95%CI=0.20-0.99). Also, sex (95%CI=1.53-6.10) and higher level of lesion (95%CI=1.31-6.15) was associated with sex life satisfaction.  When variables of sex, age and having spina bifida were examined for their association with life satisfaction, having spina bifida was associated with satisfaction with self-care ability (95%CI of odds ratio=2.52-15.61) and financial situation (95%CI=0.24-0.65), and sex was associated with sex life satisfaction (95%CI=1.04-2.73).	<b>Strengths:</b> Use of reference group randomly drawn from the general Dutch population.  <b>Limitations:</b> Absence of adolescents aged 16 and 17 yrs in the reference group, increased chance of false positives due to multiple comparisons.
Beal, Riddle, Kichler, Duncan, Houchen, Casnellie, Woodward, & Corathers 2016 <b>USA</b>	To compare youth with various chronic condition types with youth without any chronic conditions in terms of transition readiness.	Quantitative, cross-sectional  Theoretical models in self-management and social functioning (theory not specified)  Level of Evidence: IV	163 AYA (ages 12-22 yrs) ; type 1 diabetes (n=35), Turner Syndrome (n=35), autism spectrum disorder (n=35), spina bifida (n=23), AYA without any chronic conditions (n=35).	Transition Readiness Assessment Questionnaire 5.0 (TRAQ), 3-item measure adapted from Readiness for Transition Questionnaire-Teen and Transition Readiness Investigation Project for readiness to transition health care responsibility, 3-item measure adapted from the same instruments	Multiple regression, path analysis, measurement equivalent test.	<b>Differences in transition readiness according to condition</b> There was significant difference parent-child transition of responsibility, readiness to transfer to adult provider, TRAQ total score, and 3 TRAQ subscales (medications, appointments, daily living activities). Post hoc analysis showed that AYA with ASD, SB, Turner Syndrome had significantly lower TRAQ scores than AYA without chronic conditions.  <b>Conditions and TRAQ tasks (SEM results)</b> Age, family composition, health literacy and type of chronic condition was related to total TRAQ score. Gender, health literacy, type of chronic conditions were related to Parent/Child Responsibility. Different contribution patterns were observed according to condition type.	<b>Limitations:</b> Small sample size particularly for spina bifida (n=23), sample with broad age range (12-22yrs), single-center study. Condition severity and medical care participants received were not assessed. Cross-sectional design.

Author(s)	Purpose/Aim	Design	Sample	Measures	Analysis Strategy	Relevant Results	Strengths/Limitations
				for transfer readiness.			
Bellin, Dicianno, Levey, Dosa, Roux, Marben, & Zabel 2011 USA	To examine interrelationships of sex, level of lesion, self-management, community integration, and quality of life in young adults with myelomeningocele.	Quantitative, cross-sectional  Framework: Bio-neuropsychosocial model of psychosocial adjustment for emerging adults with myelomeningocele  Level of Evidence: VI	50 young adults with myelomeningocele (ages 18-25 yrs, mean=21yrs 5months); 78% Caucasian, 56% female.	Adolescent Self-Management and Independence Scale II (AMIS II), World Health Organization Quality of Life (WHOQOL-BREF), 2 questions to measure community integration (employment status, independent living)	Descriptive statistics, multivariate ANOVA, logistic regression, multivariate ANCOVA.	1. Descriptive statistics 51% earned high school degree or General Education Diploma. 8% completed college and 10% were current college students. 58% were unemployed, and 74% reported living in supervised environment.  2. Findings of multivariate analyses (1) Self-management model Level of lesion (LOL) was a significant predictor of AMIS II condition management subscale score (Roy's largest root=0.23, F=5.34) with higher mean scores for individuals with sacral LOL (5.81) than thoracic LOL (3.78). (2) Community integration: employment model Lumbar LOL (95%CI 0.004-0.78 with thoracic LOL as reference group, 0.003-0.40 with sacral LOL as reference group) and sex (95%CI 0.006-0.47) were significant predictors of employment status with males more likely to report employment than females, Good model fit was indicated (Pseudo R <sup>2</sup> : Cox and Snell=0.33, Nagelkerke=0.44). (3) Community integration: independent living model Sex (95%CI 1.96-599.27) and self-management (95%CI 1.37-11.92) were significant predictors of independent living with females more likely to report independent living. Good model fit was indicated (Pseudo R <sup>2</sup> : Cox and Snell=0.43, Nagelkerke=0.65). (4) QOL model LOL was a significant predictor of QOL (Roy's largest root=0.32, F=3.18). Social relationships subscale scores of individuals with sacral (mean=4.23) or thoracic LOL (mean=4.20) were higher than those with lumbar LOL (mean=3.37).	<b>Limitations:</b> Small sample size, recruitment from multidisciplinary clinics, cross-sectional design.
Berens & Peacock 2015 USA	To describe patient population of the Transition Medicine Clinic (TMC) established at Baylor College of Medicine in 2005 and the patients' health care needs.	Quantitative Retrospective health record review prior to July 2011.  Level of Evidence: VI	332 patients seen in TMC (mean age at first TMC visit was 22.6 yrs), 46.7% were male, The most common diagnoses included cerebral palsy (n=80), Down	Demographic and clinical data, resource and technology utilization, pediatric and adult specialist usage	Frequencies	Among 52 patients with spina bifida, technologies most commonly used were wheelchair (69.2%), urinary catheterization (69.2%), and VP shunt (67.3%). Specialties most commonly utilized included urology (59.6%), neurosurgery (28.8%) and physical/occupational therapy (28.8%). 25% had history of recurrent urinary tract infection.	<b>Limitations:</b> Single-center study, retrospective health record review

Author(s)	Purpose/Aim	Design	Sample	Measures	Analysis Strategy	Relevant Results	Strengths/Limitations
			syndrome (54), spina bifida (52), genetic conditions (50) and autism (17).				
Berry, J. G., Kusminsky, M., Foley, S. M., Hobbs, N., Queally, J. T., Bauer, S. B., & ... Weitzman, E. R. 2013 USA	To understand factors facilitating health care transition experiences for patients with SB.	Qualitative, individual interviews Level of Evidence: VI	15 individuals with SB (ages 14-28 yrs) and their parents (n=14)  5 male, 7 cognitively limited, 9 with high degree of medical complexity	Semi-structured qualitative interviews using interview guides for individual with SB and parent.	Deep reading and thematic analysis	<p><b>Factors for youth progressing toward HCT</b> Individual factors: higher cognitive functioning, strong interest in being independent from parents Family factors: exposures to parenting style that fostered independence Service factors: success negotiating primary care responsibilities with familiar adult healthcare providers</p> <p><b>Factors for youth NOT progressing toward HCT</b> Individual factors: lower cognitive ability, impaired physical ability Family factors: lack of parent awareness of medical transition needs Service factors: strong dependence on pediatric providers for care coordination, negative experiences with adult specialists perceived as not anticipating their needs</p> <p>Other relevant factors: insurance, social and life transition to adulthood first, medical transition later</p>	<p><b>Strengths:</b> Strengths and weaknesses of youth in terms of HCT were identified.</p> <p><b>Limitations:</b> Small sample size. Cognitive ability was measured as dichotomous variable. Non-English speaking families were excluded. Data on socio-economic status were limited.</p>
Betz, C., L., Smith, K., & Macias, K. 2010 USA	To compare the effectiveness of cognitive-behavioral program of Transition Preparation Training (TPT) in combination with SB management, with SB management only. The dependent variables were (1) subjective well-being, (2) role mastery, and (3) self-care practice.	Quantitative Experimental pre- and post-test design: prospective randomized controlled trial. T1: baseline, T2: 4 months later (average 4.3 months vs. control data at 4.1 months)  *TPT was a 3-module, 8-session program that involved development of an adolescent-	65 adolescents with SB (31 in experimental group, 34 in control group; ages 14-18 yrs, experimental group mean=16.19 yrs, control group mean=15.71 yrs).	Personal Adjustment and Role Skills Scale (PARS III), Community Life Skills Scale (CLSS), Denyes Self-Care Practice Instrument (DSCPI-90) Parents completed the PARS III and the parent version of the TDQ. Adolescents completed five subscales of the CLSS, the DSCPI-90© and	Descriptive statistics, linear regression model.	<p>There were no significant differences between groups of any treatment effect or treatment follow-up interaction.</p> <p>Dependency subscale of PARS III significantly increased and regularity-organization-routines subscale of CLSS significantly decreased post-treatment when the treatment and control groups were combined. However, no substantial changes in the scores either between treatments or post treatments were observed.</p>	<p><b>Strengths:</b> Prospective randomized controlled trial design. Convenience sample, but ethnically diverse.</p> <p><b>Limitations:</b> Sensitivity and specificity of the instruments used were unsatisfactory. Theoretical framework for the intervention was not presented.</p>

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		centered transition plan based on the assessment of adolescents' future goals. Level of Evidence: II		the youth version of the TDQ.			
Binks, J., Barden, W., Burke, T., & Young, N. 2007 <b>Canada</b>	(1) to identify barriers related to the transition to adult health care for CP and SB patients, (2) to identify key elements to be included in transition programs, and (3) to review empirical evidence on transition process and outcomes.	Literature review Search terms: transition, health care transition, pediatric health care, adult health care, health care access, health care use, chronic illness, special health care needs, physical disability Data sources: MEDLINE, CINAHL (1990-2006)  Level of Evidence: V	149 articles (54 discussion, 21 case series, 28 database or register, 25 qualitative, 34 survey)	N/A.	Narrative synthesis of results reported ROL. Thematic analysis identified barriers and 5 key elements that support a positive transition to adult-centered health care.	Sample included: 36% discussion studies, 17% qualitative studies, 23% survey results, 19% secondary analyses of pre-existing databases/registers, and 14% case series reports. Barriers: reluctance of child-centered health care providers, reluctance of youths, resistance of parents, lack of training and experience of adult health care providers. 5 key elements for positive transition to adult health care were: (1) preparation, (2) flexible timing, (3) care coordination, (4) transition clinic visits, and (5) interested adult-centered health care providers. Empirical evidence supporting these key elements was limited; fragmentation of services after adolescence, young adults' decline in the contact with health/social services, lack of communication between pediatric and adult-centered health care systems, families' dissatisfaction with service delivery. Therefore research that generates evidence is needed.	<b>Strengths:</b> Comprehensive literature review supplemented by empirical research reports on youths with other chronic health conditions.  <b>Limitations:</b> Methods and data analysis of source articles were unclear. Comparisons across studies were difficult due to various/unclear sampling methods, age range, and sample characteristics. No analysis by diagnosis.
Bjornson, K., Kobayashi, A., Zhou, C., & Walker, W. 2011 <b>USA</b>	To (1) assess the levels of participation in postsecondary education and employment, and (2) examine relationship of PT/OT services provided during secondary education to postsecondary education and employment in young adults with physical	Quantitative Longitudinal, secondary analysis of data from the National Longitudinal Transition Study 2 (NLTS2)  Framework: WHO's ICFDH (stated in discussion.)  Primary RQ:	990 youth with physical disabilities including CP (52%) or SB (8%) and parents/guardians. Data collection wave1: mean age=15.1yrs, range 13-17 Wave4: mean age=21.1yrs, range 19-23. *Sampling weights	Questionnaire developed for the study Most variables were measured by a single item for example dependent variables measured by 1)Any type of postsecondary education" and 2)"Worked for pay in last 2 years or currently has a paying job".	Descriptive statistics, multivariate logistic regression. All analyses were weighted with sampling weights provided by NLTS2	<b>Levels of participation at wave4:</b> 48% participated in postsecondary education in the past 12 mos; 25% attended postsecondary education at the time of survey. 43% had paid employment in the past 2 yrs; 20% were employed at the time of survey. 29% were not engaged in any employment, postsecondary education, and/or job training. <b>Logistic regression models:</b> Youth who received OT/PT services during secondary education years were 3.2 times more likely to participated in postsecondary education than those who did not receive any PT/OT services (OR=3.2). Among those who received therapy, greater self-care skills and hand functions were associated with postsecondary education (OR=4.8, and OR=1.3, respectively).	<b>Strengths:</b> Longitudinal design. Large national representative sample. <b>Limitations:</b> Secondary analysis; data were collected prior to identification of research questions. Compensations attempted by using proxy measures. Data on type and intensity of PT/OT services not available in NLTS2 dataset. Wave4 data may underestimate employment due to youth participation in

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	disabilities (orthopedic impairment, cerebral palsy, or SB).	"What is the association of PT/OT services received to levels of enrollment in postsecondary education and attaining paid employment?"  Level of Evidence: IV	performed to adjust sample distribution for key variables using known population data.	Self-care was the sum of dressing and a feeding items, both coded on a 10-point Likert scale (1 = total dependence, 10 = independent). Expressive language = average speaking and communicating items on a 4-point Likert scale (1 = speaks/communicates as clearly as other children/no trouble communicating to 4 = does not speak or communicate at all). The overall social skills variable = sum of 3 separate items querying social assertion, self-control, and cooperation		Receiving therapy services was not significantly associated with paid employment at follow-up (OR=0.44) after covariates were controlled. Among wave4 respondents, therapy and social interactions or high school diploma were positively associated with paid employment (OR=12.2 and 4.2, respectively). Among participants older than 21, receiving therapy did not increase likelihood of employment; among those older than 21 who received therapy, wheelchair use and stronger expressive language skills were positively associated with paid employment (OR = 51.4 and 5.0, respectively).	postsecondary education/ job training at the point. No analysis by diagnosis. No reliability or validity of summary variables
Buran, Sawin, Brei, & Fastenau 2004 USA	To measure the activities, beliefs and expectations, and perceived outcomes of adolescents with myelomeningocele.	Quantitative Cross-sectional, descriptive  Level of Evidence: VI	66 adolescents with myelomeningocele (ages 12-21 yrs); 58% female, 90% Caucasian; adolescents with cognitive impairment were excluded.	WeeFIM, Adolescent Decision-Making Inventory (ADI), Adolescent Self-Management and Independence Scale (AMIS), Snyder's Hope Scale, Harter Self-Perception Profile for Adolescents, Child Attitude Toward Illness Scale, A-COPE	Descriptive statistics, factor analysis and reliability analysis for the scales developed for the study.	On average, participants were hopeful (mean Total Hope score from Snyder's Hope Scale=2.9, SD=0.49) and mildly positive in their attitudes toward condition (mean Child Attitude Toward Illness Scale score=3.4, SD=0.84), and able to do activities of daily living independently (mean WeeFIM total score=5.6, SD=1.1). However, they did not achieve full range of developmental tasks, including decision-making (ADI mean subscale scores 0.75-0.98, mean total ADI score=0.87; scores below 1.0 reflecting less than average decision-making maturity), friendship (mean AAI friendship activity scale=2.8, SD=0.78), and household responsibilities (mean AAI chores scale=1.6, SD=0.30). Mean total AMIS score (3.7, SD=1.1) indicates that participants needed moderate levels of assistance with self-management tasks.	<b>Limitations:</b> Cross-sectional design, lack of normative data (some instruments being developed for the study although all with preliminary reliability and validity).

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						Participants' perceptions about their job competence (mean Harter Self-Perception Profile subscale score=2.9, SD=0.84), physical appearance (mean=2.6, SD=0.89), romantic appeal (mean=2.4, SD=0.78), scholastic competence (mean=2.4, SD=0.78), and athletic competence (mean=2.0, SD=0.85) were not positive.	
Cox, A., Breau, L., Connor, L., McNeely, P. D., Anderson, P. A., & MacLellan, D. L., 2011 <b>Canada</b>	To explore adolescent and parent perspectives on transitions to adult health care for individuals with SB and examine relationship between the perspectives and medical outcomes.	Quantitative  Level of Evidence: VI	24 patients with SB who attended adult SB clinic at an institution.	SF-36, Ambulatory Care Experience Survey, Transition of Care Survey-patient version/-parent version.	Descriptive statistics, MANOVA, MANCOVA	<b>Transition process:</b> Mean age of patients transitioning to adult health care was 26.8yrs (range 19-66). 29.2% lived independently. 62.5% wheel chair dependent. 33.3% had employment. 50% received postsecondary education (community college/university). <b>Independence and SF-36:</b> Unemployment and parent completing surveys were negatively associated with SF-36 physical role subscale scores. (p=.011 and .006, respectively). <b>Medical outcomes and SF-36:</b> Neither urological nor neurosurgical outcomes was significantly associated with SF-36 ratings. <b>ACES:</b> Medical outcomes was not significantly associated with ACES. Neither markers of independence nor medical outcomes was significantly associated with patient satisfaction with transition of care. <b>TCS (barriers and key elements derived from Binks et al.):</b> TCS-patient version scores and parent version scores were similar. Patients and parents were both satisfied with adult SB clinics; however, scores were lower than pediatric SB clinics. No other statistics were provided.	<b>Strengths:</b> Use of SF-36 and ACES, which had been validated.  <b>Limitations:</b> Low response rate (22.8%), small sample size. TCS-p, a measure developed specifically for the study, had not been validated.
Darrah, J., Magill-Evans, J., & Galambos, N. 2010 <b>Canada</b>	To understand the contribution of educational, employment, transportation and assured income service programs to successful transition to adulthood for young adults with motor disabilities.	Qualitative Interviews  Level of Evidence: VI	76 young adults (35 males, ages 20-30 yrs, 54 with CP and 22 with SB).	Semi-structured interview conducted in participants home	Content analysis	Central theme: Paradox of Services High school education programs often lead to limited future educational and career options. Employment training prepares for job seeking but not for job success or retention. Transportation options restrict employment and social choices. Assured income assures dependence rather than independence. The poverty paradox: costs are more, income is less.	<b>Limitations:</b> No analysis by diagnosis. Findings may not apply to other countries whose community services differ.
Deroche, Holland,	To develop and validate a tool to	Quantitative Instrument	174 AYA diagnosed with	Survey questions were selected	Pearson correlation	Out of 154 potential questions were selected from 4 validated surveys, 30 were included in factor analysis.	<b>Limitations:</b> Participants consisted of

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McDermott, Royer, Hardin, Mann, Salzberg, Ozturk, & Ouyang 2015 USA	measure the impact of disability on QoL in AYA with disability transitioning to adulthood.	development  Level of Evidence: VI	spina bifida (SB) (37.9%), muscular dystrophy (MD) (35.6%), or fragile X syndrome (FXS) (26.4%). 56.3% were ages 15-19 yrs, 43.7% were 20-24. 64.4% were males.	from the American Community Survey, the National Longitudinal Transition Study-2, and RAND-36.	coefficients. exploratory factor analysis, one-way ANOVA	Principal factor analysis with selection of maximum priors option was performed and the questions with absolute value of loading value being greater than 0.45 were retained. The factor analysis with Promax rotation resulted in 20 questions loaded on 5 factors: emotional health, physical health, independence, activity limitations, and community participation. Factor 1: Emotional health (4 items) Loading values 0.70-0.85, Community 0.74-0.79 Factor 2: Physical health (5 items) Loading values 0.69-0.79, Community 0.55-0.69 Factor 3: Independence (4 items) Loading values 0.54-0.76, Community 0.54-0.61 Factor 4: Activity limitations (4 items) Loading values 0.56-0.95, Community 0.60-0.91 Factor 5: Community participation (3 items) Loading values 0.69-0.80, Community 0.52-0.67  SB group reported significantly higher score on Independence subscale than FXS and MD groups.	volunteers who agreed to complete an online survey. Sample sized for some questions were too small for the factor analysis is to be performed. Lack of reliability, especially test-retest.
Dicianno, B. E., Bellin, M. H., & Zabel, A. T. 2009 USA	To examine relationships between mobility and self-management skills, psychological health, or quality of life among young adults with SB.	Quantitative Cross-sectional.  Level of Evidence: VI	61 young adults with SB (ages 18-25 yrs, mean=21.0 yrs, 60.7% female).	Hopkins Symptom Checklist-25, AMIS II, World Health Organization Quality of Life (WHOQOL)-Brief Instrument	ANOVA, Kruskal-Wallis test, chi-square/exact statistics	Full-time wheelchair users (FT) were more likely to have a history of hydrocephalus. No significant differences were found in Hopkins Symptom Checklist-25 (psychological distress) scores or in AMIS II (self-management) among 3 mobility groups (ambulators/part-time wheel chair users, FT). FT had significantly lower total WHO-QOL scores than ambulators and part-time users after controlling for hydrocephalus and motor level.	<b>Strengths:</b> Multi-center recruitment.
Galambos, N., Magill-Evans, J., & Darrah, J. 2008 Canada	To identify individual, family, and community-level variables that contribute to psychosocial maturity in the transition to adulthood for individuals with/without motor disabilities.	Quantitative Cross-sectional.  Framework: Bronfenbrenner's bioecological model of human development.  Level of Evidence: VI	74 persons with SB or CP (52 with CP, 22 with SB; 40 male, 34 female; ages 20-30 yrs, mean=25) and 72 persons without motor disabilities.	Gross Motor Function Classification System, Kaufman Brief Intelligence Test, Coping Inventory for Stressful Situations, Center for Epidemiological Studies Depression scale, Family Assessment Device Version 3, Parental Attachment	Correlations, hierarchical regression analysis	<b>Hierarchical Regression:</b> On total sample, lower IQ ( $\beta=-0.16$ ), higher levels of task-focused coping ( $\beta=0.47$ ), and lower depression ( $\beta=-0.43$ ) from Block 1 (Individual), and father's fostering of autonomy ( $\beta=0.14$ ) from Block 2 (Family) were significantly associated with greater psychosocial maturity ( $R^2=0.58$ ).  On the sample of individuals with motor disabilities, higher age ( $\beta=0.21$ ), higher task-focused coping ( $\beta=0.58$ ), and lower depression ( $\beta=-0.36$ ) from Block 1, and father's fostering of autonomy ( $\beta=0.26$ ) from Block 2 were significantly associated with greater psychosocial maturity ( $R^2=0.71$ ).	<b>Strengths:</b> Use of theoretical framework.  <b>Limitations:</b> No analysis by diagnosis. Comparison group was not matched with the motor disability group. Same data as Darrah et al. 2011 above.

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				Questionnaire fostering of autonomy subscale, Erikson Psychosocial Inventory Scale			
Grimsby, Burgess, Culver, Schlomer, & Jacobs 2016 USA	To identify barriers to transition to independent self-care in young adults with neurogenic bladder and examine which subgroups were more prepared for transition.	Quantitative, cross-sectional  Level of Evidence: VI	79 patients with neurogenic bladder referred to a transitional urology clinic.  *Of the 42 patients who completed the TRAQ (ages 18-25 yrs, mean=19.5; 60% male), 90% had SB.	Transition Readiness Assessment Questionnaire (TRAQ)	Descriptive statistics, Wilcoxon Rank Sum test	73% came to their appointment at transitional urology clinic. The most frequent reason for missed appointments was related to insurance coverage (47%). Overall mean TRAQ score was 3.0±0.89 on 5-point scale (3="I am learning to do this"). No significant differences were observed in mean TRAQ scores between sexes, ambulation status, or age groups (those ≥19 yrs vs. <19 yrs). The highest scores were observed in items related to medications, medical supplies, communication with health care providers, and household duties. The lowest scores were observed in items about health insurance coverage, payment for medications/medical equipment, obtaining financial help, and community service use.	<b>Strength:</b> Use of validated instrument (TRAQ) to measure readiness for transition.  <b>Limitations:</b> Small sample size.
Hartman, McPherson, Maxwell, & Lindsay 2018 Canada	To explore how the application of ICF-CY framework to experiences of a new transition program is considered by different stakeholder groups.	Qualitative, interviews  Level of Evidence: VI	9 youth taking part in a new transition program (ages 14-21 yrs), 11 parents, 12 YA who had not taken part in the program (comparison group) and 21 stakeholders (12 clinicians and 9 key informants)	Semi-structured interviews in person or by telephone	Qualitative thematic analysis	ICF-CY captured many key areas concerning youth's transition to adult healthcare and adult functioning. 4 domains related to body structures and functions (i.e., bowel and bladder related functions, confidence, emotional focus, sexual functions and functions related to pregnancy) and 5 domains related to activities and participation (i.e., maintaining one's health, family relationships, acquiring a place to live, productive occupations, recreation and leisure) from ICF-CY were addressed by all three participant groups.  12 environment factors and 8 personal factors that had impact on transition to adult health care were addressed by all three groups.  ICF-CY did not fully captured experiences related to transition program, independence, autonomy, and parents' roles.	<b>Limitations:</b> The ICF-CY framework was not incorporated in the research design and did not guide the transition program youth participated in. The interview guide did not reflect the elements of ICF-CY.
Heffelfinger, Koop, Fastenau, Brei, Conant, Katzenstein, Cashin, & Sawin 2008 USA	To test neuropsychological impairment, exemplified by executive dysfunction, as a risk factor in the Ecological Model of Adaptation for	Quantitative Cross-sectional  Framework: Ecological Model of Adaptation for Adolescents with Spina Bifida	43 adolescents with spina bifida (ages 12-21 yrs, mean=15.7); 56% female, 9% ethnic minority	Kaufman Brief Intelligence Test, Wisconsin Card Sorting Test, Trail Making Test, Symbol Search subtest of WISC-III, Conners' Continuous Performance	Multiple regression (hierarchical)	<b>Hierarchical Regression:</b> Age ( $\beta=0.40$ ), level of lesion ( $\beta=0.31$ ), and executive functioning ( $\beta=0.30$ ) were significantly associated with functional independence outcomes in adolescents with SB (adjusted $R^2=0.51$ ).	<b>Limitations:</b> Small convenient sample, lack of ethnic and geographical diversity, limited range of illness severity.

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	Adolescents with Spina Bifida.	Level of Evidence: VI		Test, Adolescent Decision-Making Scale, Adolescent Activity Inventory-Chores subscale, Snyder Hope Scale, Family APGAR, WeeFIM			
Heller, Gambino, Church, Lindsay, Kaufman, & McPherson 2016 <b>Canada</b>	To describe how young people with SB think about romantic and sexual relationships with partners and the impact of the condition on the relationships.	Qualitative Phenomenological design with descriptive thematic analysis  Level of Evidence: VI	11 individuals with SB (ages 16-25 yrs; 9 females).	Semi-structured interviews in person or by telephone	Thematic analysis	3 main themes were identified: (1) Importance of disclosure Participants had different ideas as to what they disclose to their partners and whether they should disclose their diagnosis.  (2) Worries around disclosure Subthemes included "timing of disclosure," "fears of rejection," and "lack of preparation for discussions about sexuality and spina bifida," lack of SB-specific sexual education (e.g. issues related to incontinence and lack of sensation)  (3) Confidence after disclosure. Some participants gained confidence after disclosing their condition; but disclosure did not always result in partner's positive response as shown in the theme of "fears of rejection."	<b>Strengths:</b> Sample with broad age range (16-25yrs), diverse gender identity and sexual orientation. Coding was conducted as a team with relevant expertise.  <b>Limitations:</b> Sample included individuals who did not have experiences of romantic and sexual relationships. Most participants were female. Study conducted in one geographic area.
Johnson, Dudgeon, Kuehn, & Walker 2007 <b>USA</b>	To examine a historical database on AYA with SB to gain better understanding of their use of assistive technology and level of community participation.	Quantitative Cross-sectional, descriptive  Level of Evidence: VI	348 AYA with SB ages 13-27 yrs from Longitudinal database maintained at Children's Hospital and Regional Medical Center, Seattle, Washington.	Assessments on demographic, physical therapy, occupational therapy, assistive technology use, and developmental status from most recent record of each individual were used. A composite variable was created to examine personal care independence.	Descriptive statistics Group differences were examined between adolescents (13-17 yrs) and young adults (18 yrs and older).	Condition: 37% of adolescents had sacral lesions. 37% of young adults had mid and low lumbar-level lesions.  Living situation: 93% of adolescents lived with one or both natural parents, whereas 75% of young adults are in the same living situation.  Education and employment: All adolescents were in some type of education. Only 50% of young adults had finished high school and 14% had finished college education or vocational/technical training. 71% of young adults were unemployed.  Assistive technology use: 52% of the whole sample used manual wheelchairs and 5% used electric wheelchairs. 35% used braces and 23% used some form of walking aids. 90% reported some kind of bowel programs and 97% reported programs for bladder management.  Personal care independence:	<b>Limitations:</b> Only limited range of data on assistive technology based on medical model were available.  Assistive technology use in school or work settings remained unclear.

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						<p>Both groups showed similar characteristics. More than 70% of the whole sample reported moderate to high independence.</p> <p>Usual Locomotion: 30% of adolescents reported ambulation without braces/aids outside home. Only 16% of young adults reported ambulation without braces/aids and 36% were wheelchair independent. 53% of young adults reported independent transportation (automobile/public).</p> <p>Participation: 72% of the whole sample reported no participation in structured activities.</p>	
Kinavey 2006 USA	To investigate how intersubjective experiences, including cultural narratives of disability, constitute self-understandings in late-stage adolescents with spina bifida.	Qualitative Phenomenological (interpretive) ethnography using in-depth interviews method  Level of Evidence: VI	11 adolescents with spina bifida who did not have a history of mental retardation (ages 18-24 yrs, mean=21); 7 females, 5 white	Longitudinal interviews (n=4) with late stage adolescents using interview guideline	Within case analysis and between case analysis of narratives from adolescents were conducted. Themes, paradigm cases, and exemplars regarding self-understanding in the adolescents were identified.	Participants were noted to claim disability in relation to identity in one of 3 ways: (a) Identity as Overcoming Disability, (b) Identity as Objectifying Disability, and (C) Identity as Integrating Disability.	<b>Limitations:</b> Small sample size (n=11).
Kinavey 2007 USA	To investigate the experience of disability and its attached meanings in relation to self, world, and other for late-stage adolescents with spina bifida.	Qualitative Phenomenological (interpretive) ethnography using in-depth interviews method  Level of Evidence: VI	11 adolescents with spina bifida who did not have a history of mental retardation (ages 18-24 yrs, mean=21); 7 females, 5 white (note same sample/data as in 2006 study but different research question)	Longitudinal interviews (n=4) with late stage adolescents using interview guideline	Themes and exemplars of narratives on adolescents' self and experiential world were identified by conducting within case and between case analysis. Used ethnograph for data storage, retrieval, analysis and theme building	The theme described by the participants in common in relation to the biopsychosocial developmental effects of living with the disability of the condition was "Experiencing self as dissimilar other."	<b>Limitations:</b> Small sample size (n=11).
Levy, Wynd, & Carachi 2014 UK	To examine the experiences of service users in	Both qualitative and quantitative	Focus groups: 1st session by Skype with 2	Semi-structured framework with core questions	Recurring themes were identified.	Recurring key themes identified in the focus group data included issues concerning "communications," "respect" to the young person, "person-centered care," and "choice	<b>Strengths:</b>

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	the transition process in Scotland.	data were collected. Qualitative: focus groups Quantitative: survey (as complimentary data)  Level of Evidence: VI	families, 2nd face-to-face session with 12 parents.  Survey: 7 AYA with SB (ages 12-25 yrs) on the SSBA National database and 13 parents.	was used for focus group discussions.		and control" related to children's decision-making.  Survey results showed that many participants felt that the aspects of "level of control" and "choice" were not yet relevant to them. However, many of those who participate in 'negotiating the system' and those who were expected to make decisions felt powerless. (Only summary of survey results were provided.)	Quantitative data were collected to supplement focus group data.  <b>Limitations:</b> Low survey response rate (12%).
Lindsay, Fellin, Cruickshank, McPherson, & Maxwell 2016 <b>Canada</b>	To explore youth and parent experiences of a new transition model for youth with spina bifida, compared to the experiences of young adults with spina bifida who did not participate in the model.	Descriptive, qualitative  Level of Evidence: VI	32 participants (9 youth, 11 parents, 12 young adults)	Semi-structured interviews in person or by telephone	Thematic analysis	The analysis revealed variables that could be either a benefits or barriers to transition including: Structural factors - [Continuity and availability of care; Location of clinics; Organization of care [Different models of care]; Relationship factors [Trust/rapport with HCP; Family involvement]; Personal factor [Transition readiness and self-care] Most youth and parents felt supported by pediatric providers and benefitted from gradually transferring responsibility from parents to youth, reported experiencing challenges, including lack of support from primary care providers and lack of clarity about the new model. Many of those who did not take part in the model reported receiving some transition-related thought support from pediatric specialists, parents, and in some cases, primary care providers. But, they also reported experiencing gaps in their continuity of care and had unmet needs for support with employment, relationships, finances, and housing.	<b>Strengths:</b> Large sample size for qualitative study using in-depth interviews.  <b>Limitations:</b> Low response rate. Mailed information package to youth and parents (n=64) and young adults (n=128), and 32 participants took part in the study. Most youth had not transferred out of pediatric health care. Limited ethno-cultural backgrounds of participants.
Lindsay, McPherson, & Maxwell 2017 <b>Canada</b>	To explore characteristics of school-work transitions for youth with SB.	Qualitative  Level of Evidence: VI	9 youth and 12 YA with SB 11 parents and 12 clinicians (Youth ages 14-21 yrs and parents invited to participate in a new transition model and youth aged 19-25 and parents not invited)	Semi-structured interviews in person or by telephone	Qualitative content analysis	13 youth and young adults had characteristics categorized as "non-adaptive" transitions, 10 "approaching" transitions and 6 in "adaptive" transition stage.  Factors enabling school-work transitions: professional support from pediatric and adult health providers for transition to post-secondary education or employment, social support (e.g., peer mentoring, support from parents), and school accommodations and support from teachers. Barriers to school-work transitions: Lack of education and information on finances, housing and transportation Challenges of coping with SB: Lack of support, challenges finding work, discrimination and stigma at school and work.	<b>Strengths:</b> Perspectives of youth, parents, and health providers were compared.  <b>Limitations:</b> Secondary analysis of a larger study that did not focus on school-work transitions. "Parents" included only mothers.
Liptak, Kennedy, &	To examine transition	Quantitative, prospective	130 individuals with SB	Survey on 6 categories based	Descriptive statistics, chi-	At wave 1 (mean age=15.3 years), 94% were enrolled in a regular school and 4% were in special education	<b>Limitations:</b> Classification as SB made by

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Dosa 2010 USA	outcomes and identify factors affecting social participation in adolescents and young adults with SB.	cohort study with 4-year follow up (wave 1 in 2001, wave 2 in 2003, wave 3 in 2005).  Framework: The World Health Organization's International Classification of Functioning, Disability, and Health framework  Level of Evidence: IV	representing a weighted sample of 5171 individuals (mean age at wave1=15.3 yrs, range 13-17; 44% female, 62% White, 19% Latino, 16% African-American)	on WHO's ICF framework: (1) health condition, (2) body function and structure, (3) activities, (4) participation, (5) environment, and (6) personal.	square test, ANOVA, logistic regression	setting. 47% reported being teased, and 24% reported being bullied at school. 21% were classified to have learning disability (LD). At wave 3 (mean age=19.3 years), 79% lived with parents, 76% were employed or attended post-secondary school within the past 2 years, and 36% had a job in the past 12 months. The proportion of individuals reporting excellent general health and those reporting fair/poor health significantly changed from wave 1 to 3 ( $\chi^2=761$ , $df=1$ ). The results of logistic regression with variables reflecting participation from WHO's ICF model from wave 3 as dependent variables (DVs) yielded various patterns of associations with independent variables from wave 1. Among the independent variables, being Latino was significantly negatively associated with the participation variables from wave 3 (95% CI of odds ratio=0.007-0.03 with DV "Participated in volunteer/community service," 0.17-0.28 with DV "Spent time with friends or going on dates."). Also, not speaking English at home was negatively associated with the dependent variables (English spoken at home 95% CI of odds ratio=5.0-16.7 with DV "Competitively employed or attending post-secondary school, 4.08-9.04 with DV "Shopping/hanging out at the mall or coffee shops or just driving around with someone." ).	school district and confirmed by parents was used to identify the sample. No data were collected on shunted hydrocephalus and cognitive ability. No control group was available.
Mahmood, Dicianno, & Bellin 2011 USA	To examine relationships among self-management, patient assessment of health care services, preventable conditions, and health care utilization for young adults with SB.	Quantitative, cross-sectional  Framework: The Chronic Care Model (CCM)  Level of Evidence: VI	38 individuals with myelomeningocele receiving care from 5 multidisciplinary SB clinics around U.S. (mean age=22.2 yrs, 52.6% female, 78.9% Caucasian).	Questionnaire packet including demographic questions, recent medical information, and health care usage. AMIS II Patient Assessment of Chronic Illness Care (PACIC)	Descriptive statistics, multiple regression	<b>Occurrence of medical outcomes:</b> UTI median=2, range 0-20 in the last 3 years, pressure ulcers median=0, range 0-5 in the last year, ER visits median=0, range 0-3 in the last year, and hospitalization median=0, range 0-20 in the last 3 years.  <b>Regression models:</b> No history of shunting ( $t=-3.814$ ), lower education level ( $t=-3.227$ ), lower AMIS scores ( $t=-2.500$ ), and higher employment status ( $t=2.593$ ) were significantly associated with UTIs requiring antibiotics (adjusted $R^2=0.774$ ). Higher education level ( $t=2.372$ ) and higher motor level ( $t=2.325$ ) were significantly associated with pressure ulcers (adjusted $R^2=0.378$ ). Lower AMIS scores ( $t=-2.242$ ) and higher number of ulcers ( $t=2.903$ ) were significantly associated with hospitalizations (adjusted $R^2=0.544$ ).  PACIC scores: For most of the subsets the median scores were 3 or 4 on the 5-point scale. The median score was lowest (1) in	<b>Limitations:</b> Causality cannot be established due to cross-sectional nature of the research design. The sample consisted of individuals receiving care from multidisciplinary SB clinics, who may have had higher cognitive function than other cohorts being able to provide informed consent.

Author(s)	Purpose/Aim	Design	Sample	Measures	Analysis Strategy	Relevant Results	Strengths/Limitations
						the subset asking about encouragement to go to a class or group to help cope with SB. Total PACIC score was not significantly associated with any outcome variables.	
Mann, Royer, Turk, McDermott, Holland, Ozturk, Hardin, & Thibadeau 2015 USA	To compare emergency room visit (ER) and inpatient hospital utilization (IP) for AYA with spina bifida transitioning to adult health care to comparison group using South Carolina administrative data.	Quantitative Retrospective cohort study  Level of Evidence: IV	Spina bifida group: 695 cases of spina bifida (41.0% male, 59.0% female; 55.4% ages 15-19 yrs, 44.6% 20-24 yrs) Comparison group: 1390 cases (41.0% male, 59.0% female; 54.6% ages 15-19 yrs, 45.4% 20-24 yrs)	Total ER rate and IP rate, cause-specific rate of ambulatory sensitive conditions and other condition categories	Multivariable generalized estimating equation (GEE) Poisson models were used to make comparison between the groups.	ER and IP use rates for AYA with spina bifida were significantly higher than for the comparison group (ER use 0.83 visits per person-year vs. 0.18, IP use 0.28 visits per person-year vs. 0.03). IP hospitalizations were due to skin conditions, musculoskeletal conditions, nervous system conditions, respiratory conditions, and genitourinary conditions.  Among cases with SB, young adults (20-24 yrs old) used ER at higher rates than adolescents (15-19 yrs old) due to all ambulatory care sensitive conditions (ACSC) (incident rate ratio=1.36). Specifically, urinary tract infections was significantly more common among young adults as a reason for ER visits (IRR=1.74). No significant difference was observed in the rate of IP use between young adults and adolescents.	<b>Limitations:</b> Only billing data were collected; clinical record was not studied.  <b>Strengths:</b> Large sample size, state-wide data collection.
Østerlund, C., Dosa, N., & Arnott Smith, C. 2005 USA	To examine how young adults with SB and their parents interact with their medical records. GT approach was used to analyze (1) who is involved in record keeping, (2) how the information is stored, (3) what information is kept and shared among the different constituencies and (4) when patients and parents need information.	Qualitative Grounded theory Focus groups and structured interviews  Level of Evidence: VI	6 young adults (4 male, 2 female, ages 18-21 yrs), 6 family members (4 mothers, 2 fathers) and 1 home nurse.	Focus groups and structured interviews with questions concerning if and how young adults with SB interact with their medical record during transition to adult health care.	Two stage analysis using Grounded Theory. Data coding initially using NVIVO	Mothers play a central role in the medical record management. The parent-maintained home based records served as a linking pin in a heterogeneous healthcare information environment. The records tended to be organized as time-lines. Parents believed that they were the only source of entire health care history of the young adults. Young adults and parents supported the idea of having access to medical records on-line, and emphasized that emergency highlighted their need for complete and accessible medical record.	<b>Strengths:</b> Experience with and responsibility related to medical records is important component of transition experience.  <b>Limitations:</b> Sample not representative of the population (highly motivated parents and young adults with little cognitive impairment).
Ridosh, M., Braun, P., Roux, G., Bellin, M., & Sawin, K. 2011 USA	To explore 3 dimensions of transition experience: (1) self-management, (2) independence,	Qualitative Narrative inquiry  Framework: Ecological	10 young adults (ages 18-25 yrs; 7 with myelomeningo cele, 3 with	Individual interviews using interview guide	Narrative Interpretive analysis. Rigor procedures detailed. Results from two groups	3 themes that captured the dimensions of transition experiences emerged: (1) struggling for independence, "My parents should have made me do it sooner." (2) limiting social interactions and experiences with	<b>Strengths:</b> Use of theoretical framework.  <b>Limitations:</b> Semi-structured interview questions on independence,

Author(s)	Purpose/Aim	Design	Sample	Measures	Analysis Strategy	Relevant Results	Strengths/Limitations
	and (3) the role of inner strength in the pursuit of independence.  To explore condition-related needs and life skills required during transition.	Model of Secondary Conditions and Adaptation (Sawin & Thompson, 2009; Sawin et al., 2003)  Level of Evidence: VI	lipomyelomenin gocele)		were similar and reported as single sample.	stigma, "the looks," and (3) building inner strength, "	inner strength, and self-management were used, which may have restricted the participants responses. Sample was only from one site of data collection (a teaching hospital in Midwest metropolitan area). Sample ethnic diversity was limited.
Ruck, J., & Dahan-Oliel, N. 2010 <b>Canada</b>	(1) To determine youths's readiness for adulthood, (2) to measure their perception of teen-centeredness of the care received at Shriners Hospital for Children (SHC), and (3) identify areas for improvement in the transition program.	Quantitative, Descriptive  Level of Evidence: VI	36 individuals with SB (ages 14-21 yrs; 18 male, 18 female) attending interdisciplinary Myelodysplasia clinic at SHC in Montreal	Young Adult Transition Assessment with subdomains of staying healthy, managing your own health care, being independent, emotional health, school and work, community resources, and finding adult health care providers. Giving Youth a Voice (GYV-20) based Measure of Processes of Care and reduced by Canadian investigators	Descriptive statistics	Transition outcomes: (yes/ no format, although subdomains are proposed only items are reported.) 89% reported that they had family doctor. 65% reported being able to describe their health condition and manage health care independently. 47% reported not being responsible for making medical appointments or not keeping medical records. 94% reported being partially or completely independent with self-care. 69% did not have driver's license. 50% reported doing chores around the house. 78% reported regular attendance at schools. Almost half reported currently volunteering or having paid jobs.  Satisfaction with services: Mean scores of GYV-20 ranged between 6.00 and 6.77 (on 7-point scale). Mean scores for subscales ranged between 6.41 and 6.69.	<b>Strengths:</b> Descriptive of transition outcomes and AYA evaluation of transition program. GYV-20 with good reliability and validity data  <b>Limitations:</b> Young Adult Transition Assessment has no reported reliability or validity. Generalizability is limited because of convenience sample recruited at a hospital. Research questions focused only on the evaluation of transition services delivered at a specific health care facility (SHC).
Sawin, K. J., Rauen, K., Bartelt, T., Wilson, A., O'Connor, C., Waring, W. P., & Orr, M. 2015 <b>USA</b>	To conduct an initial evaluation of the SB Transition Project (SBTP) from the adolescent/young adult and family perspective.	Qualitative  Framework: Transition Care Model and Ecological Model of Secondary Conditions and Adaptation in SB	40 individuals with SB (24 AYA and 16 parents representing 28 families)  AYA: ages 18-35 yrs, 50% female, 92% Caucasian Parents: ages	Individual qualitative interviews conducted separately for each participating AYA and parent 2-3 weeks after initial transfer.	Thematic analysis. Parent and EA data were analyzed separately but due to the similarity of the results the two samples were combined and reported as family themes unless	6 themes included: "Positive experience," "establishing trust," "unexpected benefits," "communication," "potential worries," and "suggestions for improvement."	<b>Strengths:</b> Use of theoretical framework.  <b>Limitations:</b> Those with less positive perceptions may not have participated. Convenience sampling; those who did not have access to health services in urban setting were difficult to participate. Only initial findings of model

Author(s)	Purpose/Aim	Design	Sample	Measures	Analysis Strategy	Relevant Results	Strengths/Limitations
		Level of Evidence: VI	40-63 yrs, 88% female		there were unique findings by group.		intervention program. Longitudinal study will be needed.
Sawin, K., Bellin, M., Roux, G., Buran, C., & Brei, T. 2009 USA	To describe the experiences of self-management in adolescent women with SB.	Qualitative  Framework: Ecological Model of Adaptation in SB (Sawin et al., 2002; Sawin, Buran et al., 2003) Independent Living Model (Dejong, 1979), ICF model (ICF, WHO, 2001)  Level of Evidence: VI	31 adolescent women with SB (ages 12-21 yrs); 97% non-Hispanic white; 87% had hydrocephalus	Individual interviews using guided interview guide	Content analysis used to identify domains, themes, subthemes and exemplars	3 themes and 7 subthemes emerged on the dimensions of self-management experiences during transition: (1) Opportunities to engage in self-management activities -- knowledge, skills, and aspirations, (2) dance of individuation -- parental impact on self-management, and (3) advocacy within self-management-- confronting discrimination and stigma.	<b>Strengths:</b> Use of theoretical framework.  <b>Limitations:</b> Sample ethnic diversity is limited (primarily Caucasian). Sample consisted only of women.
Shalaby, Gibson, Granitsiotis, Conn, & Cascio 2015 UK	To evaluate Adolescent Transition Urology Clinic (ATUC) established in 2009 and to assess adolescents' perspectives on transition process and care.	Quantitative  Level of Evidence: VI	26 adolescents and young adults (ages 15-28 yrs) completed ATUC service. 19 adolescents completed satisfaction questionnaire. 22 individuals (85%) had neurogenic bladder; 14 of them had SB.	2 online urological transition questionnaires: Care Transition Measure 15 (CTM-15), Transition of Care Experience (TCE)	Descriptive statistics (frequencies)	Of 30 adolescents and young adults attending ATUC over the study period, 26 (87%) completed transition to adult urology clinic. Only one adolescent (5%) thought that the ATUC service was not beneficial. 87% would recommend the ATUC to other adolescents. 74% thought that written information would have been useful. 47% thought that 18 years was appropriate age for transition. 74% thought that written information would have been useful and 21% considered adult hospital as an inappropriate environment for treating young adults. After attending ATUC, 11% patients thought their preferences were not met. 16% were not confident that they could take care of their health.	<b>Strengths:</b> Use of validated instrument (CTM-15)  <b>Limitations:</b> Single-center study, small sample size, mixed sample with complex urological conditions, no analysis by diagnosis was conducted.
Smith, Macias, Bui, & Betz 2015 USA	To examine adolescents' reasons for participating in a spina bifida transition intervention study.	Qualitative  Level of Evidence: VI	25 adolescents (11 from treatment group, 14 from control, ages 14-20 years) who participated in the Transition Preparation Training Program (TPT), ideas as to why youth declined, recruitment suggestions for	5 survey questions asking the reasons for participating in the Transition Preparation Training Program (TPT), ideas as to why youth declined, recruitment suggestions for	Frequencies Content analysis for coding	38% of the sample participated due to the desire to learn more about their condition and 28% for altruistic purposes to help other patients. 33% speculated that unwillingness to participate would be because the adolescents were shy, embarrassed, or emotionally unable to participate. 64% responded that research incentive did not affect their decision.	<b>Limitations:</b> Sample consisted of participants of one intervention study.

Author(s)	Purpose/Aim	Design	Sample	Measures	Analysis Strategy	Relevant Results	Strengths/Limitations
				future studies, and whether the offer of research incentive affected their decision to participate in the study.			
Snow-Lisy, Yerkes, & Cheng 2015 USA	To review the evidence on urological management of spina bifida from prenatal stage to adulthood.	Literature review Search terms: spina bifida, spinal dysraphism, bladder. Data sources: MEDLINE, EMBASE, & PubMed (-December 2014).  Level of Evidence: V	437 articles were identified to potentially address issues related to management of pediatric patients, long-term outcomes, and QoL.	N/A.	Synthesis of literature addresses individual's challenges with urological management and impact on independence as well as the type of urological management	Individuals with SB have more difficulty compared to typical emerging adults due to physical issues, such as obesity and sexual dysfunction, mental health issues, such as depression and cognitive challenges, and social issues, including loss of support from school system etc.  The role of pediatric urologists in improving independence, self-care, sexual function, and successful transition to adulthood and adult health care and the importance of a structured transition programs is emphasized.	<b>Strengths:</b> Includes individual's physical and psychosocial challenges with urological management in adulthood  <b>Limitations:</b> Focus is on the urologist role.
Squiers, Lutenbacher, Kaufman, & Sharp 2017 USA	This quality improvement project aimed to identify barriers and opportunities for successful transition from pediatric to adult health care for a single clinic	Quality improvement study with quantitative and qualitative components. Health Utilization model provided an overall conceptual framework  Level of Evidence: VI	Pediatric patients (N = 16) with spina bifida ages 14-19 yrs and a family member recruited from the Spina Bifida Clinic at a large regional medical center.	A standard guide, including select questions from the National Survey of Children with Special Health Care Needs along with inquires specific to the population and clinic setting, were used for each interview.	Descriptive statistics. Content analysis was used to analyze data from open-ended questions and identify common themes.	Quantitative findings: 62.5% of the participants had not had a preventable health checkup in the previous year. Only 25% reported previous conversation with their pediatric providers regarding transition to adult health care. Supports identified as beneficial in transition planning included meeting adult providers prior to transition (93.8%), a written transition plan (68.8%), and possessing a copy of the transition policy (68.8%).  Qualitative findings: Parents of children with greater intellectual and physical disability reported increased worry surrounding transition and not receiving information on transition when compared to parents of higher functioning children. Parents desired information about what they could expect from adult providers, what services would be covered, what types of issues they needed to look for, and how to contact resources.	<b>Strengths:</b> Both quantitative and qualitative findings are provided.  <b>Limitations:</b> Single-center study, small sample size, limited gender and ethnic diversity of sample (69% male, 87.5% Caucasian).
Stephany, Ching, Kaufman, Squiers, Trusler,	To (1) assess the readiness of spina bifida patients and their parents regarding	Quantitative  Level of Evidence: VI	15 SB patients (ages 14-31 yrs, mean age 19.1) and 18 parents.	Questionnaire developed for the study.	Fisher's exact test for comparisons between patients and parent responses, chi-	88.9% of parents had thought about transition process while only 40% of patients did. 22.2% of parents and 40% of patients never wanted to transition. Previous history of stones was significantly associated	<b>Limitations:</b> Small sample size, questionnaire not validated.

Author(s)	Purpose/Aim	Design	Sample	Measures	Analysis Strategy	Relevant Results	Strengths/Limitations
Clayton, Thomas, Pope, Adams, Brock, & Tanaka 2015 USA	transition process and (2) identify factors associated with successful transition.				square test for comparisons between transitioned group and nontransitioned group	with transitioning to adult health care. Of the 97 patients identified to be eligible for transition, 33 patients (34%) had either been transitioned or had plan to meet an adult urologist.	
Strömfors, Wilhelmsson, Falk, & Höst 2017 Sweden	To investigate children's and adolescents' experiences of living with SB, their social and emotional adjustment, their thoughts about becoming independent adult.	Qualitative  Level of Evidence: VI	8 young persons with SB ages 10-17 yrs	Semi-structured interviews, the Beck Youth Inventories (BYI) to assess social and emotional problems.	Qualitative content analysis	Three themes were identified: (a) being a person with SB, (b) everyday living as a person with SB, and (c) preparing for life as an adult with SB.  Finding from responses to BYI: Emotional and social problems were most prominently indicated among participants with milder physical disability (i.e., participants who were able to walk without aid).	<b>Strengths:</b> BYI provided data on social adaptation problems supplemental to interviews.  <b>Limitations:</b> Small sample size, wide age range (10-17 yrs),
Summers, Elliott, McAdams, Oottamasathien, Brant, Presson, Fleck, West, & Myers 2014 USA	To identify urologic problems and needs of patients with SB during transition to adult health care.	Quantitative Retrospective health record review  Level of Evidence: VI	65 patients from SB clinic at the universities of Utah and Minnesota (April 2011-April 2012; mean age 30.6 yrs).	Health record review for methods of current bladder management, urologic problems, time to presentation after referral from pediatric care, compliance with needed screening, and required surgical and medical interventions. Transition time was calculated from the date of last urologic follow-up to first SB adult clinic visit.	Fisher's exact test, chi-square test	85% reported urologic problems at the time of SB clinic visit. Common problems included urinary incontinence (52%), recurrent urinary tract infections (34%), catheterization troubles (12%), and calculi (9%). 97% required some form of urologic intervention, including diagnostic, surgical and medical.  No statistically significant difference was found in transition times between individuals with any urologic problems and those without.	<b>Limitations:</b> Retrospective chart review, 18% of patients had no information for calculating transition time, data collection from two study sites without standardized tool
van der Toorn, Cobussen-Boekhorst, Kwak, D'hauwers, de	To examine specific needs of adolescent urologic patients regarding	Quantitative, descriptive.  Level of Evidence: VI	58 individuals with neuropathic bladder due to SB, exstrophy	Questionnaire based on nationwide "On Your Own Feet Ahead"	Descriptive statistics	49% of the sample had neuropathic bladder due to SB. Among pre-transfer group, knowledge on urological disease and related issues was reported as adequate except medication side effects. Majority had knowledge about preventive measures, however, 40% reported that	<b>Limitations:</b> Mixed sample consisting of individuals with SB, exstrophy and other chronic bladder disorders. No analysis by

Author(s)	Purpose/Aim	Design	Sample	Measures	Analysis Strategy	Relevant Results	Strengths/Limitations
Gier, Feitz, & Kortmann 2013 <b>Netherlands</b>	independence and transition.		or other chronic bladder disorders and parents of post-transfer group in the Netherlands. *55 pre-transfer patients (mean age=20, 56% male) and 3 post-transfer patients (ages 18-29, 67% male).  Response rate=73%.	Collaborative and adjusted and supplemented for use with urological patients.		they only knew "more or less" about knowledge on prevention of urinary tract infections. Open-ended question answers indicated that participants appreciated long contact and personal bond with pediatric urologist; they viewed building relationship with adult urologist as a barrier. Approaching post-transfer group was not successful in this study.	diagnosis. The authors created some variables, such as knowledge about diseases and related issues, by simply grouping questions and calculating Cronbach's alpha.
van Staa, A., Jedeloo, S., van Meeteren, J., & Latour, J. 2011 <b>Netherlands</b>	(1) To map experiences with transfer to adult care of young adults with chronic conditions, and (2) to identify recommendations for transitional care from the perspective of young adults, parents, and healthcare providers.	Qualitative  Level of Evidence: VI	24 young adults after transfer (ages 15-22 yrs; diagnoses hemophilia 3, DM 3, SB 6, CHD 3, CF 3, JRA 3 and sickle cell anemia 3)  24 parents and 17 healthcare providers	Semi-structured interviews using interview guide	Thematic analysis	Experiences and views of YAs, parents, and HCPs mainly overlapped and were condensed into 4 core themes: Regarding moving to adult health care, (1) leaving pediatric care is a logical step, (2) Transition is complicated by cultural gaps between pediatric and adult services, and Regarding recommendations, (3) better patient and parent preparation, and (4) better organization and communication between pediatric and adult health care.	<b>Limitations:</b> Possible selection bias; some conditions showed lower response rates than others. Possible overrepresentation of those with severer conditions. Only hemophilia department offered structured transition program.
Warschawsky, Kaufman, Schutt, Eviitts, & Hurvitz 2017 <b>USA</b>	To examine the associations between generic and condition-specific health self-management and levels of adaptive behavior in 2 groups of transition-age youth with congenital neurodevelopmental condition	Quantitative, Cross-sectional  Level of Evidence: VI	43 adolescents/young adults with cerebral palsy (CP) and 36 with spina bifida/myelomeningocele (SBM), ages 13-29, mean age 18.96 (4.77), 51.9% female	Transition Readiness Assessment Questionnaire (TRAQ) [child]; Kennedy Krieger Independence Scales-Spina Bifida (KKIS-SB) [parent/guardian]; The Adaptive Behavior Assessment System (ABAS-II)	Descriptive statistics, Chi-squares, bivariate correlations, hierarchical linear regressions	There were significant group differences in condition-specific health self-management, including lower KKIS-SB Initiation of Routines and KKIS-SB Prospective Memory scores in the group with SBM. Group differences in generic health self-management [ABAS-II] and adaptive behavior [KKIS-SB, TRAQ] were not significant. There were significant differences in the correlations between health self-management instruments and ABAS-II composite scores. KKIS-SB subscale scores correlated significantly with ABAS-II composite scores in both groups.  The results of hierarchical linear regression with the whole sample showed that TRAQ subscale scores were	<b>Limitations:</b> Some KKIS-SB items related to bowel and bladder management were inapplicable to AYA with CP. Hierarchical regression analyses were not conducted separately with each diagnostic group.

Author(s)	Purpose/Aim	Design	Sample	Measures	Analysis Strategy	Relevant Results	Strengths/Limitations
				[parent/guardian] r		not significant predictors of ABAS-II composite scores in final models. KKIS-SB Initiation of Routines was significantly associated with all ABAS-II Composite scores ( $\beta=-.55, -.41, -.50$ and adjusted $R^2=.45, .32, .35$ with Conceptual, Social and Practical Composite scores, respectively).	
Woodward, J., Swigonski, N., & Ciccarelli, M. 2012 USA	To assess health, functional characteristics, and health care service needs of youth and young adults attending comprehensive, noncategorical transition program.	Quantitative, Descriptive  Level of Evidence: VI	87 individuals with chronic conditions attending noncategorical transition program, the Center for Youths and Adults with Conditions of Childhood in Indianapolis (ages 11-22 yrs; 54% male, 36% CP, 10% SB, 17% Developmental delay/Down synd., 9% autism) and their parents.  Response rate=44%.	Self-administered survey developed for the study. Questions assessing health care needs and access to needed services were adapted from the NS-CSHCN and the National Health Interview Survey.	CI, Chi-square test, t-test	Youth health status was poorer than the results of 2007 National Survey of Children's Health. Only 41% or parents reported excellent or very good health status.  69% of participants needed assistance with personal care; 91% with routine needs (e.g., eating, bathing, dressing etc.) and 59% used assistive devices. 75% were in school; 85% lived with parents.  The services most needed were dental care (95%), routine preventive care (93%), prescription medications (93%), and specialty care (78%). Compared with 2005-2006 National Survey of Children with Special Health Care Needs, parent reported higher needs for all services except mental health care and tobacco/substance use counseling.  Unmet needs were highest for physical, occupational, and speech therapy (26%). 43% reported at least one unmet health need.  No other statistics were provided.	<b>Limitations:</b> Functional limitation and health needs relied on parent proxy reports. (Youth response rate was only 27% possibly due to impairment.) Inability to determine effectiveness of the program due to absence of control group. No reliability or validity on survey instrument
Young, N., Barden, W., Mills, W., Burke, T., Law, M., & Boydell, K. 2009 Canada	To explore specific issues in transition to adult-oriented health care in a Canadian context.	Qualitative  Level of Evidence: VI	15 youths (ages 14-18 yrs) and 15 adults (age 24-32 years) with disabilities (13 with CP, 9 with SB, 8 with acquired brain injuries in childhood/ABlc ) and their parents (i.e.. 30 pairs).	Semi-structured individual interviews	Transcribed data entered into NVivo. Observational and reflexive notes were examined. Constant comparative method was used to identify themes related to transition. Themes were explored to determine if any were unique to diagnosis, stage	6 key themes, including 4 challenges and 2 solutions were identified. Those themes occurred across ages and diagnostic groups. Challenges/Barriers: 1. Lack of access to health care (78% of participants discussed.) 2. Lack of professional knowledge (56%) 3. Lack of information provided (55%) 4. Uncertainty regarding the transition process (60%) Solutions: 1. More information throughout transition process (52%) 2. More support throughout transition process (50%)	<b>Strength:</b> identified challenges in transition. Evaluated separately by group but then combined due to similarity. Discussion of similarities across those with different disabilities.  <b>Limitations:</b> Small sample size. Omits the optimal transition years 18-24.

Author(s)	Purpose/Aim	Design	Sample	Measures	Analysis Strategy	Relevant Results	Strengths/Limitations
Young, N., McCormick, A., Mills, W., Barden, W., Boydell, K., Law, M., & ... Williams, J. 2006 <b>Canada</b>	To present a comprehensive picture of health and life outcomes of both youth and young adults with CP, SB, and AB/c.	Quantitative  Level of Evidence: VI	<p>290 individuals with cerebral palsy, SB, or acquired brain injury in childhood recruited from 6 children's rehabilitation centers (CRCs) across Ontario, Canada.</p> <p>Youth cohort ages 13-17.9 yrs and adult cohort ages 23-32.9 yrs. Response rate=40.2%. (1,762 individuals received invitation letter, 709 responded, 352 consented, and 290 completed questionnaire packets.)</p> <p>Sample included 40 youth and 13 adults with SB.</p>	Items about education, living situation etc. from National Population Health Survey, Health Utilities Index, Assessment of Quality of Life, Health Assessment Questionnaire	of transition, gender, severity, or region of residence.  t-test, Pearson's correlation, Chi-square test	<p><b>Total Sample Findings:</b> 95% of youth and 61% of adults were living with their parents. 23% of youth and 55% of adults were employed. 60% of youth and 42% of adults reported very good to excellent health status; adults with SB reported lowest. 17.4% of youth reported transitioning to adult health care system, and the mean age of transition was 12.5 (SD=5.2) yrs. 86% of adults reported transitioning to adult health care, and the mean age of transition was 18.0 (SD=2.64) yrs.</p> <p><b>SB Findings:</b> 100% of youth and 61.5% of adults were living with parents/siblings. 38.5% of adults finished university-level and 15.4% college level education. 30.8% of adults were working full-time, and 23.1% part-time. 25% of youth reported their health status as excellent, 40% very good, and 30% as good. For adults, proportions of those who reported good or fair/poor were 38.5% respectively.</p>	<p><b>Strengths:</b> Analysis by diagnostic subgroup. Participants with the lowest health scores were reported by adults with SB.</p> <p><b>Limitations:</b> Sample recruited only from Ontario CRCs (However there were 6 of these from across Ontario).</p>

**Manuscript #B. The Transition Experiences of Adolescents and Emerging Adults  
with Spina Bifida: A Synthesis of Literature Published in Japanese Journals**

**Abstract**

**Background:** In Japan, the prevalence of spina bifida (SB) is steadily increasing. As life expectancy of the individuals with SB extends, transition to adulthood has become an important issue. To explore the state of the science on experiences of Japanese adolescents and emerging adults (AEA) with SB and to identify gaps in the current body of literature, a synthesis of research literature about AEA with SB published in Japanese journals was conducted.

**Methods:** A Japanese healthcare research database, *Igaku-chuo-zasshi* (Japan Medical Abstracts Society), MEDLINE, and CINAHL were searched using the following keywords: “spina bifida” or “myelomeningocele,” “young adult\*,” “emerging adult\*,” “adolescen\*,” and “Japan.” Articles published during the years between 2004 and 2016 as well as historical articles were included for review.

**Results:** The final sample consisted of 22 studies. Five themes were identified: (a) challenges of self-management, (b) health care needs, (c) struggling to achieve social adaptation, (d) threats to mental health, and (e) complexities in quality of life.

**Conclusion:** Knowledge gaps in the existing literature on AEA with SB in Japan and implications for future research and for nursing practice were discussed. More research is needed to develop and provide effective services to meet complex health care needs of the AEA with SB during transition.

*Keywords:* spina bifida, Japan, adolescents, emerging adults, transition

Spina bifida (SB) is a complex congenital condition characterized by neural tube impairments. Although the prevalence of SB has been on a declining trend worldwide (Rosano et al., 1999; Williams et al., 2005), in Japan, it steadily increased from 1.72 per 10,000 births between 1974 and 1978 to 5.93 in 2010 (The Center of International Clearinghouse for Birth Defects Surveillance and Research, 2012). The survival rate among infants born with SB are improving continuously worldwide (Wong & Paulozzi, 2001), thus making transition to adulthood one of the most important issues for individuals with SB and their families.

Adolescents with SB are faced with various challenges when transitioning to adulthood both in Japan and in the rest of the world. These challenges may be related to their physical and cognitive disabilities, such as limited mobility, neurogenic bowel and/or bladder, and learning disability. In Japan, researchers are increasingly expanding their scope for research to include person centered approach including the psychosocial aspects of transition to adulthood for adolescents with a chronic health condition (Fujita, 2010a). A synthesis of this growing body of Japanese literature is an important step to identify what is known and what the gaps in knowledge are for Japanese adolescents and emerging adults (AEA) with SB, and develop interventions to address the needs of the population.

### **Purpose**

The purpose of this chapter is to explore the transition experiences of Japanese AEA with SB. Critiques of the research design and samples of the studies are also provided. In addition, gaps in the current body of literature on the experiences of Japanese AEA with SB are identified. Implications for future research and for nursing practice are discussed.

### **Methods**

## Identification and Selection of Studies

The articles for this review were selected from literature on experiences of Japanese AEA with SB written either in Japanese or about Japanese AEA written in English. Such literature was identified by searching *Igaku-chuo-zasshi* (Japan Medical Abstracts Society), which is one of the leading Japanese databases in health care, MEDLINE, and CINAHL. The primary keywords used in the search included: “spina bifida (spinal dysraphism as MeSH term in MEDLINE and CINAHL),” “myelomeningocele,” “young adult\*,” “emerging adult\*,” and “adolescen\*.” “Japan” was added as a search term when literature search was conducted using MEDLINE or CINAHL. Historical articles published before 2004 that were identified by the authors as crucial to understanding the state of the science of Japanese AEA with SB were included.

The inclusion criteria for this review were research studies: (a) with Japanese AEA or parent as participants, (b) written in Japanese or English in Japanese journals, (c) published during the years 2004 and 2016 and (d) that focused on the experiences of Japanese AEA during transition into adulthood, including but not limited to self-management, psychosocial issues, and beliefs/ experiences related to the illness and disability. The exclusion criteria were: (a) articles published before 2004, (b) articles that were not peer-reviewed research reports, (c) dissertations, (d) single case studies, (e) studies that addressed only epidemiological aspect of SB, and (f) studies that addressed only specific physical or neurocognitive functions, such as neurological function, urinary tract function, renal disorder, gait analysis, orthotics, associations with other illnesses (e.g., enuresis), latex allergy, pressure ulcer management, sexual function, fetal screening, and prenatal prevention.

In this review, an adolescent was defined as a young person aged 10-17 years. This age range was chosen as it represents the developmental stage characterized by changes toward independence and identity exploration (WHO, 2014). An emerging adult was defined as a person aged 18-29 years who starts to accomplish more independence and explore different possible choices in adult life. (Arnett, 2015).

One hundred and seventy articles were identified through *Igaku-chuo-zasshi*. Twenty-eight articles were excluded due to duplication. Twenty-one articles were also identified by MEDLINE and two by CINAHL. Six articles were excluded due to duplication. After the titles and abstracts of the remaining 159 articles were reviewed, 147 articles were excluded because they did not meet the inclusion criteria. Ten articles were identified by studying reference lists and added to the sample. Consequently, 22 articles were included in the final sample. All of the studies included for review were identified through *Igaku-chuo-zasshi* or by hand search. Of the 22 articles, 21 were written in Japanese and one in English (see Figure 2.2). Two articles on quality of life (QOL) in individuals with SB (Yoshida & Iwaya, 2000) and on menstrual hygiene management in women with SB (Douki & Iwaya, 2008) were included in spite of the fact that they were not peer reviewed because the former measured QOL with the instrument that has been internationally tested for reliability and validity and the latter addressed important issues concerning self-management in women with SB. The sample in both studies included AEA with SB.

### **Analysis Strategies**

After reviewing the full articles, essential information was extracted and entered in an evidence table to facilitate the interpretation of the data. The table includes information

regarding study purpose, design, framework, sample, measures, analysis, findings, and limitations (See Table 2.2). The guideline for assessing level of evidence developed by Melnyk and Fineout-Overholt (2011) was used to evaluate the quality of the studies.

### **Findings and Discussion**

The analysis of the literature generated the following themes: challenges of self-management, health care needs, struggling to achieve social adaptation, threats to mental health, and complexities in QOL. The results of the analysis and critique are reported below.

#### **Challenges of Self-Management**

Eight studies addressed self-management behaviors practiced either by Japanese AEA with SB or by their parents (Domae, Date, & Sakuda, 2014; Hashizume et al., 2003; Hori, Narama, & Yamauchi, 2002; Kihara & Suzuki, 2008; Koike, 2007, 2011; Noda et al., 2013; Take, 2004). The behaviors addressed in the literature included managing six components (a) medications (Hori et al., 2002), (b) bowel programs, including enema/cleansing enema (Domae et al., 2014; Hashizume et al., 2003; Hori et al., 2002; Kihara & Suzuki, 2008; Koike, 2007; Take, 2004), (c) neurogenic bladder, including clean intermittent catheterization (CIC) (Domae et al., 2014; Hori et al., 2002; Kihara & Suzuki, 2008), (d) skin care (Hori et al., 2002; Kihara & Suzuki, 2008), (e) dressing (Hori et al., 2002), and (f) menstrual hygiene management (Noda et al., 2013).

The majority of adolescents with SB reported using CIC to manage their neurogenic bladder. More than 70% of adolescents with spina bifida in Japan had bowel incontinence (Take, 2004). The most prevalent method of bowel management was digital removal of feces (Domae et al., 2014; Kihara & Suzuki, 2008), followed by enema, cleansing enema and use of

medications (Domae et al., 2014; Kihara & Suzuki, 2008). Three studies described the degrees of independence and family involvement in self-management behaviors at different ages or grades (Hori et al., 2002; Kihara & Suzuki, 2008; Take, 2004). Authors of those studies identified that individuals with SB often started to practice CIC on their own or under parental supervision early in elementary school (Hori et al, 2002; Kihara & Suzuki, 2008). More than half of adolescents at the ages between 10 and 12 and 90% of adolescents in junior high school were responsible for managing CIC (Hori et al., 2002). On the other hand, parental management was prevalent until higher grades in elementary school or middle school ages in bowel management, including digital removal of feces (Hori et al, 2002; Take, 2004), enema, cleansing enema, and skin care (Hori et al., 2002). Data on when the AEA assumed complete responsibility for bowel program self-management was not available.

In a quasi-experimental clinical study using a small sample the study participants reported that they became less anxious about soiling especially at school after starting cleansing enema (Hashizume et al., 2003). Only two studies described the psychological and social process in which AEA with SB acquired bowel-management behaviors (Koike, 2007, 2011). Individuals with SB developed bowel management skills after experiencing multiple failures, and the issues related to bowel management increased their awareness of difference from others (Koike, 2007). They felt uncertain about the effectiveness of medical treatment suggested by health care providers and wanted more reliable solutions (Koike, 2007). Also, in another qualitative study, women with SB attempted to obscure their difference from others and developed their own coping methods, such as modulation of the level of disclosure about illness and its self-management, selective use of information, and use of their own coping strategies that

fit with their lifestyles and illness status, in order to maintain self-esteem (Koike, 2011).

Menstrual hygiene management is one major aspect of self-management for AEA women with SB. Two studies explored issues related to menstrual hygiene management in this population (Douki & Iwaya, 2008; Noda et al., 2013). Study findings on the women's attitude toward menstruation have been mixed. Approximately 60% of women with SB aged 11 to 39 years reported that they viewed menarche as a positive event (Noda et al., 2013) while only 30 % viewed it positively in the other study that used a sample of women with SB from adolescence to menopausal ages (Douki & Iwaya, 2008). The proportion of the women who reported menstrual cycle irregularities ranged from 35 to 39% (Douki & Iwaya, 2008; Noda et al., 2013). It is important to note that 90% of the women with SB from adolescence to menopausal ages performed menstrual hygiene care independently (Douki & Iwaya, 2008). Approximately 20% of the women practicing CIC reported that they had difficulties in performing CIC during the menstrual period (Noda et al., 2013) and 30% experienced problems in bowel management due to premenstrual diarrhea and/or constipation (Douki & Iwaya, 2008). Other premenstrual symptoms related to bowel and bladder management included cloudy urine, bladder incontinence, bladder pain, urinary frequency, and difficulty of urination (Douki & Iwaya, 2008). In addition, mothers of the women reported concerns about the women's menstrual hygiene and related issues, such as symptoms linked to menstruation, menstrual cycle, and urinary tract infection (Noda et al., 2013).

Other aspects of self-management, such as consulting health professionals when necessary and understanding medical information were addressed in only one study (Domae et al., 2014). Eighty percent of the emerging adults and adults (age 18 to 64) with SB were able to

ask physicians questions and state their own opinions, but only half of the participants understood what types of medical equipment they used (Domae et al., 2014). On the other hand, only 36% of the participants understood their medications, including their effects and side effects; likewise, only 46% of the participants responded that they were able to consult health professionals about reproductive health and sexual issues (Domae et al., 2014).

### **Health Care Needs**

Seven studies explored health care needs of Japanese AEA with SB and their families and services provided to meet the needs (Date & Ito, 2003; Date, Ito, & Numata, 2008; Date, Ito, Numata, & Matsuoka, 2005; Domae et al., 2014; Oi, Sato, & Matsumoto, 1996; Nishi & Okada, 2008; Suzuki, Matsuno, Noda, & Adachi, 2014). Bowel management and management of neurogenic bladder were the most common needs expressed by individuals with SB and their families (Date & Ito, 2003). Pressure ulcers may be another health issue of importance for this population. It is the most common reason for hospitalization in individuals with SB over the age of 18 (Domae et al., 2014).

The vast majority of individuals with SB visited two or more clinics at a time to see specialists because they had multiple problems associated with their condition (Date & Ito, 2003). These problems varied depending on the types of the condition. Individuals with spina bifida occulta, the mildest form of SB that may be unrecognized for years after birth because the skin on the back is intact and a small sinus track on the lower back may be the only early sign of neurological impairment, tended to have needs and concerns about bowel and bladder management, urinary tract infection, employment, education, familial difficulties, and sexual difficulties. However, those with myelomeningocele, the most severe form of SB where the

vertebrae do not fuse, the skin is open at birth with meninges protruding and who need surgery to close the lesion in the first days of life, had needs or concerns about bowel/bladder management, cognitive status, education, shunt status, urinary tract infection, employment, and mental health (Date & Ito, 2003; Oi et al, 1996). In both cases, however, many of these concerns involved issues related to transitions to adult life and to adult health care.

One solution for alleviating patients' difficulties due to such multiple clinic visits may be a multidisciplinary program that provides individuals with SB with comprehensive health care services (Date et al., 2005, 2008). At the time of this review, only one multidisciplinary SB program has been reported in the literature in Japan (Date et al., 2005, 2008). The most common need that brought the individuals and their families to the multidisciplinary clinic was related to life adaptations in school-related dimensions, followed by transition to adult health care and to independent living, and bowel/bladder management (Date et al., 2005, 2008). The patients and families who received the multidisciplinary services tended to positively evaluate the problem solution provided at the clinic (Date et al., 2005, 2008). Only one study examined the types of the health care facilities the emerging adults and adults with SB visited in childhood and at the age of 18 and older (Domae et al., 2014). More than 60% of the participants were seen in health care facilities specializing in pediatric health care in childhood. Among these individuals, 40% continued to be seen in the same health care facilities at age 18 and older (Domae et al., 2014).

One study examined concerns regarding sexual and reproductive health issues among young women with SB (Suzuki et al, 2014). The study revealed that approximately 60% of female college students/working adults had anxiety and worries about sexual and reproductive

health issues (Suzuki et al., 2014). Concerns regarding pregnancy and childbirth were the most common for the college students/working adults. Approximately 30% of the parents of the female college students/working adults with SB also had concerns related to sexual and reproductive health issues; the proportion of the parents who had concerns was significantly higher than that of the women with SB themselves (Suzuki et al., 2014).

The other problem addressed in the literature related to health care needs of individuals with SB was parental perceptions of their children's multiple disabilities. The process in which mothers of children and adolescents attending regular schools understood their children's illness was described in a grounded theory study by Nishi and Okada (2008). The process consisted of two stages: the mothers understood that their children had bowel and bladder function impairments first, and they next became aware of the children's learning issues as their children's social relationships expanded in late elementary school grades (Nishi & Okada, 2008). The mothers experienced disappointment and sorrow especially when the learning issues became obvious. The findings indicated the importance of support for mothers of children and adolescents with SB attending regular schools. Especially important was understanding the impact of the condition on the psychological status of the mothers (Nishi & Okada, 2008).

### **Struggling to Achieve Social Adaptation**

Two studies explored issues related to social adaptation of Japanese AEA with SB. Most individuals had some difficulties in learning and social skills, such as difficulties in math skills, memory-making, and group activities (Kihara & Yamaguchi, 2010). A qualitative study on experiences of individuals with SB who had received education both in regular schools and in special education settings revealed that school-age children and adolescents with SB were

viewed as unique and unusual in a negative sense by other students without disabilities due to their special needs when they entered regular schools. This stigma may have induced school bullying (Koike, 2008). On the other hand, adolescents with SB felt that they were “not special” (Koike, 2008, p.32) in special education settings and found the settings comfortable (Koike, 2008). These adolescents reported significant difference in life adaptation experiences between regular schools and special education settings (Koike, 2008). This situation surrounding school-age children and adolescents with SB may be due to cultural beliefs about disability and education system for individuals with disability in Japan, where inclusive policies and environments at schools are still less prevalent than the international community (Aoki, 2011). People with disabilities are often hidden by their families from their neighbors (Kayama, 2010). Their education has been traditionally provided only in special education settings separate from regular classrooms. Although a new system for special education was introduced in Japan in 2007, it is still difficult for students with disability to receive education designed based on the principles of disability rights (Kayama, 2010).

### **Threats to Mental Health**

Four studies assessed mental health of Japanese AEA with SB (Fujita, 2010b, 2014; Fujita & Sueda, 2009; Ono & Inaba, 2008); however, mixed findings were reported. QOL scales for children and adolescents often include a mental health domain (Solans et al., 2008). In the above-mentioned QOL study by Ono and Inaba (2008), the anxiety and distress subscale may have measured an aspect of the mental health status of Japanese adolescents with SB, revealing that adolescents with SB had significantly higher QOL subscale scores in this domain than healthy controls (Ono & Inaba, 2008). This finding may be due to the fact that the sample in

this study consisted of Japanese adolescents with SB younger than 15 years, who lived in the social environment where they were able to receive care and assistance from parents and other adults easily (Ono & Inaba, 2008).

In a study guided by Adler's individual psychology framework, however, the mean score of inferiority feelings produced by others' perceptions was significantly higher and the mean score of subjective well-being was significantly lower in emerging adults with SB than in typically developing controls (Fujita, 2010b). Interestingly, however, there was a positive relationship between inferiority feelings caused by lack of confidence and subjective well-being in emerging adults with SB, indicating that inferiority feelings did not undermine their psychological well-being (Fujita & Sueda, 2009). Also, the other study using the same framework showed that subjective well-being of Japanese emerging adults and adults with SB was significantly associated with the number of friends with SB and feelings of belonging to the society and feelings of trust (Fujita, 2014). The findings may suggest the role of social participation in improvement of subjective well-being of emerging adults with SB (Fujita, 2014).

### **Complexities in Quality of Life**

Three studies examined QOL in individuals with SB (Kihara & Suzuki, 2008; Ono & Inaba, 2008; Yoshida & Iwaya, 2000). Of these, Yoshida and Iwaya (2000) employed SF-36 to measure QOL in individuals with spinal lipoma and Ono and Inaba (2008) examined QOL in school-age children and adolescents with SB using a life satisfaction scale developed by Japanese researchers (Nakamura et al., 2002) and adapted for use with children and adolescents with SB. The other examined variables related to illness, disability, self-management, learning, education, and employment as QOL indicators (Kihara & Suzuki, 2008).

The findings of comparisons of QOL scores in individuals with SB with that in typically developing controls have been inconclusive. The subscale scores of SF-36 in the domains of role limitation related to physical health problems and role limitation related to emotional problems in individuals with spinal lipoma were significantly lower than normative scores from typically developing controls (Yoshida & Iwaya, 2000). The physical functioning subscale score in individuals with spinal lipoma or myelomeningocele was significantly associated with their paralysis states (Yoshida & Iwaya, 2000). On the other hand, comparison of QOL in adolescents with SB or healthy controls revealed that overall QOL mean score reported by adolescents with SB was significantly higher than that of typically developing controls (Ono & Inaba, 2008).

Also, among adolescents with SB, those who practiced CIC for bladder management had lower QOL scores in the dimension regarding anxiety and distress than those who did not (Ono & Inaba, 2008); however, it was unclear which caused the psychological problems, CIC itself or the bladder impairment that required CIC. Approximately 25% of individuals with SB reported that they were unsatisfied with the places in their schools where they practiced CIC (Kihara & Suzuki, 2008). This dissatisfaction with the school environments may be associated with the lower QOL in adolescents practicing CIC. QOL research with AEA with SB still appears to be in its early phase. Further accumulation of research will be necessary in this area.

### **Comparison between the Finding and the Literature on Transition Written in English**

The comparison between the findings of the two reviews of literature on transition for AEA with SB to adulthood revealed several similarities and differences. The qualitative studies on the experiences of individuals with SB revealed that psychosocial challenges, such as stigma,

discrimination, or bullying (Koike, 2008; Ridosh et al., 2011; Sawin et al., 2009) were experienced by individuals with SB both in the Northern America and in Japan. Further, these challenges were associated with perception of difference between others and self in both countries (Kinavey, 2006; Koike, 2011), suggesting that those challenges are common across countries and cultures.

The most significant difference was that there was no experimental study to test the effectiveness of transition programs for AEA with SB in Japan while one such experimental study has been conducted and another study appears to be under development in the U.S. (Betz et al., 2010; Sawin et al., 2014). The development of effective transition programs for these AEA is imperative in both regions; however, there seemed to be a considerable difference in the progress of research.

Finally, in Japan, few studies have addressed experiences of transition into adulthood for AEA with SB although the importance of successful transition to adult life is growing (Koike, 2011; Koike, 2008). Moreover, two of these studies had somewhat limited scope of inquiry, i.e., the process in which the individuals with SB acquired bowel management skills (Koike, 2008) and the experiences of attending regular and special needs schools (Koike, 2008). It appears that research in European and North American countries tended to be conducted from more holistic perspectives than in Japan.

## **Quality Assessment**

### **Study Design and Sample**

Of the 22 studies reviewed, eighteen were descriptive or correlational, quantitative studies and four were qualitative studies. No experimental study was found except for the

quasi-experimental study that examined patients' evaluations of the effects of colonic irrigation (Hashizume et al., 2003). All the studies were rated at level 6 (evidence from qualitative or descriptive study) using the rating system developed by Melnyk and Fineout-Overholt (2010).

There were only three studies in which the samples consisted of emerging adults with SB aged 18 to 35 (Fujita, 2010b, 2014; Fujita & Sueda, 2009). In some studies (Hashizume et al., 2003; Ono & Inaba, 2008; Take, 2004) the samples consisted of school-age children with SB that included adolescents. The majority of the studies, however, included individuals with SB in wide age ranges and two studies (Hori et al., 2002; Nishi & Okada, 2008) included only mothers of children with SB in elementary and junior high schools. Although sample sizes varied considerably, ranging between eight (Koike, 2008) and 1,112 (Kihara & Suzuki, 2008), studies that used large-scale samples tended not to focus on AEA with SB but to include individuals in wide age ranges.

### **Knowledge Gaps and Implication for Future Research**

This analysis of research studies on Japanese AEA with SB highlighted future research directions. First, few studies specifically focused on AEA with SB; rather, the studies reviewed often used samples with a wide age range (Date & Ito, 2003; Date et al., 2008; Douki & Iwaya, 2008; Kihara & Suzuki, 2008). More studies specifically addressing adolescence and emerging adulthood in individuals with SB will be needed in Japan since individuals in these developmental phases have unique problems and needs related to transition into adulthood and adult health care. Also, the studies reviewed often relied primarily on data reported by parent proxy (Date & Ito, 2003; Date et al., 2008; Kihara & Suzuki, 2008). It is important to shed more light on the perspectives of AEA with SB themselves.

Second, some studies assessed QOL of Japanese individuals with SB; however, there was only one study that used an instrument that measured AEA's perception (rather than function) and could enable international comparisons. Use of QOL scales developed to facilitate international comparisons, such as WHOQOL-BREF (Skevington, Lotfy, O'Connell, and WHOQOL Group, 2004), would reveal the current status of QOL of Japanese AEA with SB and domains where Japanese AEA have different scores than those in other countries. Use of generic QOL instruments would also promote comparisons between QOL scores of individuals with SB and those with other conditions. Such comparisons may be useful in improving health care for Japanese AEA with SB.

Third, although the importance of making successful transition to adult life is increasing among AEA with chronic conditions, few studies addressed their experiences, psychological or social processes in which Japanese AEA with SB made transition into adulthood (Koike, 2007, 2008, 2011). More studies will be needed in this area to fully describe the transition for this population. Such knowledge will contribute to the development of effective health care services aimed at facilitating transition.

Finally, although the theory of emerging adulthood (Arnett, 2015) offers a useful framework for research on Japanese AEA with SB, it may be insufficient for understanding their developmental transition interacting with the chronic health condition. In addition to the basic emerging adulthood theory, a theory specific to transition in AEA with the chronic conditions in Japan may need to be developed.

### **Implication for Nursing Practice**

One prominent issue for AEA with SB in transition is that they visit clinics with a broad

array of needs from specific health care needs related to self-management behaviors to educational and/or vocational needs. There is a growing awareness of the need for multidisciplinary health care programs for individuals with SB in Japan (Date, Ito, Numata, & Matsuoka, 2005; Date, Ito, & Numata, 2008). Nurses caring for this population will need to participate in the development of such programs from nursing perspectives to meet those individuals' holistic needs. In addition, as in the cases of other chronic conditions, transition to adult health care is a key issue for AEA with SB. Coordination of care for these individuals during transition will be an issue of priority for nurses since they have highly complex health and psychosocial needs.

Studies on menstrual hygiene management in Japanese adolescent and emerging adult women with SB revealed that some symptoms and problems related to menstruation affected their self-management behaviors and transition outcomes especially in bowel and bladder management (Douki & Iwaya, 2008; Noda et al., 2013). In order to facilitate optimal self-management outcomes both in menstrual hygiene and in other areas, such as bowel and bladder management, nurses and nurse-midwives need to pay more attention to these women's menstrual hygiene care.

Adaptation to regular school settings has been a major challenge for Japanese children and adolescents with SB (Koike, 2008). Nurses caring for school-age children and adolescents need to provide support not only to facilitate their self-management behaviors in those schools but also to adjust relationships of the children and adolescents with other students and teachers. These roles of nurses may become increasingly important because special education system is changing in Japan due to the special education reform implemented in 2007 aimed at providing

special education services based on individual educational needs (Kayama, 2010). The findings of QOL studies also have implication for nursing. For example, the lower QOL subscale score in psychological domain in adolescents with SB who practiced CIC (Ono & Inaba, 2008) indicates that nurses working with adolescents with SB may need to pay more attention to school environments where the adolescents perform self-management behaviors.

### **Conclusions**

Twenty-two studies on the experiences of Japanese AEA with SB were reviewed. The findings indicated the following: (a) Self-management was studied from several different perspectives. The process in which individuals with SB developed bowel management behaviors was described in qualitative studies. (b) AEA with SB had a variety of health care needs. One solution to the complexity of their health care needs may be the development of multidisciplinary health care programs. (c) Findings have been mixed on QOL of AEA with SB in comparison with their peers. (d) Adolescents with SB often experienced challenges in life adaptation especially in learning- and school-related dimensions. (e) Research findings regarding mental health of individuals with SB have been mixed. In one study a QOL subscale score of adolescents with SB in psychological domain was higher than that of the control group without SB, whereas in other studies AEA with SB reported lower subjective well-being than those without SB. Overall, more research is needed to develop and provide effective services to meet complex health care needs of Japanese AEA with SB.

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Figure 2.2.

Flow diagram of identification and selection of studies on experiences of adolescents and emerging adults with spina bifida published in Japanese journals.

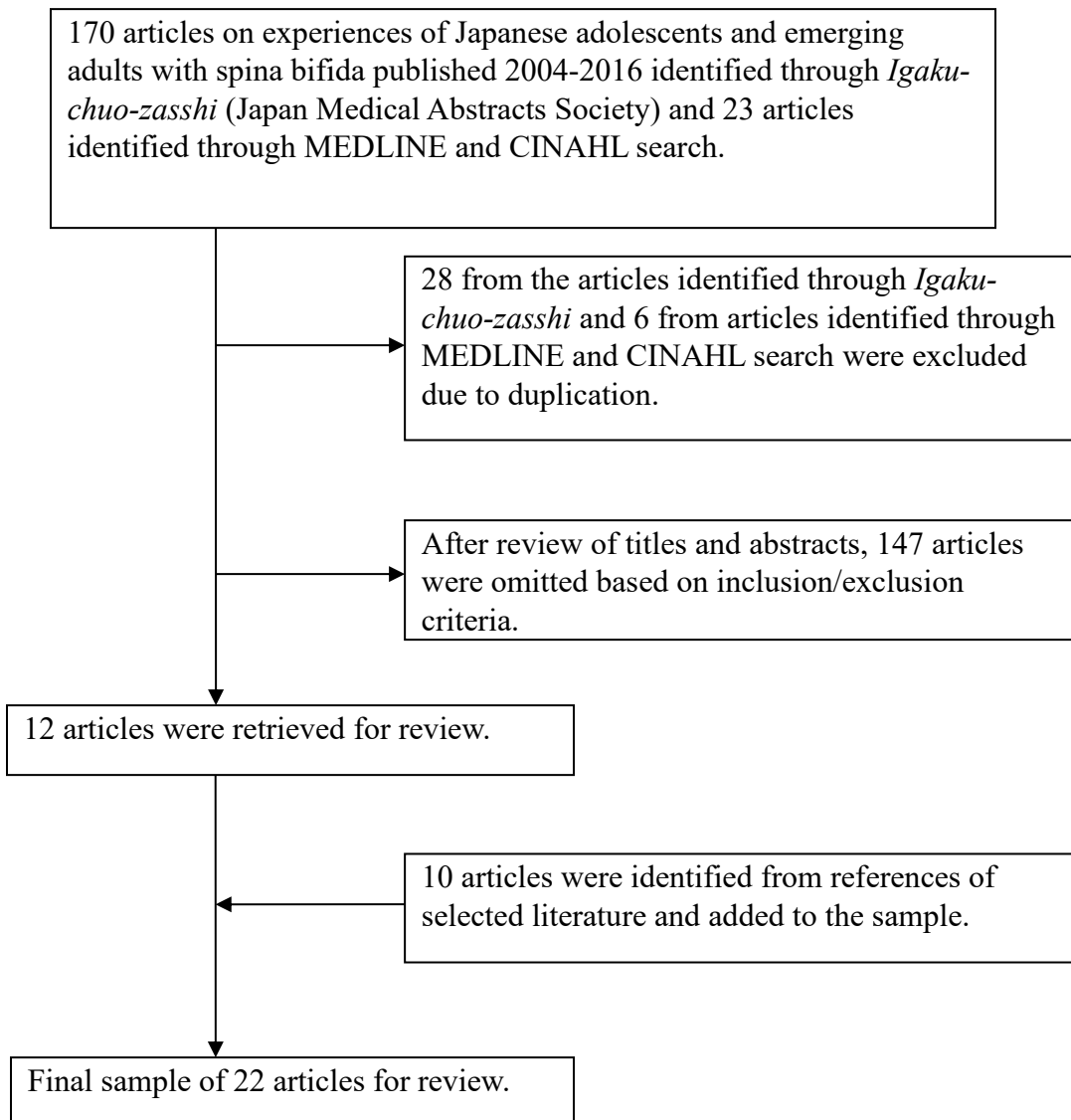


Table 2.2.

*Studies on Experiences of Adolescents and Emerging Adults with Spina Bifida Published in Japanese Journals*

Author(s)	Purpose/Aim	Design	Sample	Measures	Analysis	Relevant Results	Strengths/Limitations
Date, H., & Ito, C. 2003 <sup>a</sup>	To examine neurological and social conditions of individuals with SB	Quantitative, Descriptive, Review of medical records and survey	100 patients with myelomeningocele (ages 6 months-28 years) and 60 patients with spinal lipoma (ages 10 months-27 years)	Spina Bifida Neurological Scale for neurological condition	Descriptive statistics	27% of the patients with SB were not ambulatory while all the patients with spinal lipoma were community ambulators. 61% of the patients with SB had IQ scores above 80 when starting school. Multiple problems associated with the conditions appeared to be reasons for patients to see specialists during a clinic visit. Patients with myelomeningocele tended to have various concerns including shunt status, cognitive status, UTI, bowel/bladder management, education, and employment. Spinal lipoma patients had concerns related to UTI and bowel bladder management.	<b>Limitations:</b> Age range not limited to adolescents and young adults. No demographic data were given. No statistical tests were used. Sample consisted of SB patients from one institution.
Date, H., Ito, C., Numata, O., & Matsuoka, M. 2005 <sup>a</sup>	To explore desirable long-term management of SB based on the data on present condition of SB patients at a multidisciplinary clinic.	Quantitative Descriptive Survey	48 patients at a multidisciplinary SB clinic (ages 5-7 and around 18 years; these age ranges represent starting of school and transition to independent living)	Survey created by authors	Descriptive statistics	<b>Purpose for clinic visit</b> 39.6% of the patients visited the clinic with concerns on adaptation to school life. 29.1% visited with concerns on transition to adulthood, including independent living and transition to adult health care. <b>Questionnaire results</b> 73.7% reported that they were satisfied with the level of problem solving at the multidisciplinary clinic. Reasons for unsatisfactory problem solving included patients' and parents' consciousness of transition issues that has not been enhanced and lack of understanding among people surrounding the patients. 73.7% had their primary care physicians; however, 64.3% of those families reported that the primary care physicians had little knowledge about SB.	<b>Limitations:</b> Age range not limited to AEA adults. Survey without reliability and validity was used. No clear demographic data were given. Sample consisted of SB patients from one institution.

Author(s)	Purpose/Aim	Design	Sample	Measures	Analysis	Relevant Results	Strengths/Limitations
Date, H., Ito, C., & Numata, O. 2008 <sup>a</sup>	To evaluate services delivered at SB multidisciplinary clinic.	Quantitative, Descriptive	95 patients with SB (3 months-28 years, 82 with myelomeningocele, 9 with spinal lipoma, 4 with other types) and families	Survey created by authors	Descriptive statistics	21.1% visited the clinic with concerns related to bowel/bladder management and independent living; 36.8% with concerns on adaptation to school life; and 22.1% with concerns related to transition to adulthood. 42.9% reported that the services at the clinic completely solved their problems; 28.6% reported that most of their problems were solved.	<b>Limitations:</b> Age range not was limited to AEA. No clear demographic data were given. Sample consisted of SB patients from one institution. Survey was not tested for reliability or validity. Findings were very similar to Date et al., 2005.
Domae, Y., Date, H., & Sakuda, K. 2014 <sup>c</sup>	(1) To examine present situations of the social life and self-management of adults with spina bifida. (2) To discuss self-management strategies desired to develop during childhood to lead a fulfilling social life.	Quantitative, Descriptive	249 individuals with SB (ages 18-64 years; 126 males, 123 females) who belong to Spina Bifida Association of Japan (SBA-J). Response rate was 40%.	Questionnaire created by the authors to examine demographic background, social life, self-management, consultation behavior, and health care support received during childhood.	Descriptive statistics, Chi-square test	43% of the participants worked full-time, 8% part-time, and 12% attended vocational programs while 15% were unemployed and 9% were students. 80% of the participants practiced urinary catheterization for bladder management. Approximately 50% used diapers and incontinence pads. Out of the 200 individuals who performed catheterization, 91% did it by themselves. For bowel management, 32% of the participants used digital removal of feces and 29% used medications. Only 21% practiced cleansing enema. 60% of the individuals who practiced enema and/or cleansing enema performed the procedures by themselves. 59% of the participants needed assistive mobility devices. 35% of the participants experienced pressure ulcer after high school graduation. 42% had history of hospitalization after high school graduation; pressure ulcer was the most common reason. 80% of the participants responded that they were able to ask physicians questions and state opinions and 73% responded that they knew the symptoms that warrant consulting a physician while only 46% responded that they were able to consult with health professionals about pregnancy, childbirth, or sexual issues and concerns. 62% of the participants were seen in health care facilities specializing in child health care or in pediatric specialties in childhood. Among these individuals, 40% continued to be seen in the facilities specializing in child health care or in pediatric specialties at age 18 and older.	<b>Strengths:</b> Relatively large sample size.  <b>Limitations:</b> Age range was not limited to AEA. Sample consisted of members of SBA-J.

Author(s)	Purpose/Aim	Design	Sample	Measures	Analysis	Relevant Results	Strengths/Limitations
						29% had life-style related conditions, such as obesity and hypertension.	
Douki, K., & Iwaya, T. 2008 <sup>c</sup>	To describe current situation of menarche, menstrual cycle, symptoms, menstrual care in women with SB.	Quantitative, Descriptive	190 women with SB (53 women aged 18-20, 128 aged 21-45, 9 aged 46-59 yrs)	Survey created by authors	Descriptive statistics, Chi-square test	The average age of menarche was 12 years (range: 9-18). Premature menarche (before 10 years) occurred in 9 women; delayed menarche (after 15 years) in 12 women. 123 women had regular menstrual cycles. 147 women had premenstrual symptoms, such as irritability, low back pain, cramping, depressive feelings, and headache. In addition, 55 women had difficulty in bowel management due to premenstrual diarrhea/constipation. Some reported urinary and bladder changes, such as cloudy urine, bladder pain, incontinence, and urinary frequency. 161 women experienced symptoms during menstrual periods, such as low back pain, cramping, fatigue, and irritability. 175 women (92%) practiced menstrual hygiene management independently. 58 women viewed their menarche positively while 105 women viewed negatively.	<b>Limitations:</b> Age range not limited to AEA. Survey without reliability and validity was used. Not peer reviewed.
Fujita, Y. 2010 <sup>b</sup>	To examine how inferiority feeling and social interest influence mental health of young adults with SB.	Quantitative, Descriptive, cross-sectional	30 young adults with SB (age 18-35 yrs) and 40 healthy controls (age 19-29 yrs)	Kyodotaikankaku-shakudo [Social Interest Scale], Rettokan shakudo [Inferiority Feeling Scale], Kaitei Happiness of Being shakudo [Happiness of Being Scale-Revised]	Factor analysis, ANOVA, t-test, multiple regression	Comparisons of the mean scale scores between groups showed that inferiority feelings score was significantly higher in young adults with SB than in healthy controls ( $t=-1.69$ , $p<.10$ ), and happiness (subjective well-being) was significantly lower in those with SB ( $t=1.87$ , $p<.10$ ). None of the variables measured with Social Interest Scale or Inferiority Feeling Scale was significantly associated with happiness (subjective well-being) ( $R^2=.38$ ). Responses to open-ended questions on future prospects revealed that young adults with SB explicitly reported their interest in independence and independent living while healthy controls did not.	<b>Strengths:</b> Focused on EA Use of standardized measuring instruments. The authors performed principal factor analysis with Promax rotation to test whether the instruments can be used with both individuals with SB and healthy controls. Use of inferential statistical tests. <b>Limitations:</b> Small sample size with convenience sampling. No reliability data
Fujita, Y. 2014 <sup>b</sup>	To identify psychosocial factors that affect subjective well-being in young adults with SB.	Descriptive, Correlational	56 young adults with SB (age 19-37 yrs)	Scale of Social Interest, Happiness of Being Scale-Revised (both developed by Japanese	Multiple regression with the scale score of the Happiness of Being Scale-Revised and its subscales	Results of multiple regression analyses showed that: (1) Number of friends with SB ( $\beta=.40$ ) and feelings of belonging to society and trust ( $\beta=.63$ ) explained 58% of the variance in Happiness of Being Scale-Revised scale score, (2) Marriage status ( $\beta=.17$ ), number of friends	<b>Strengths:</b> Use of standardized measuring instruments. Regression analysis was conducted. Use of inferential statistical tests.

Author(s)	Purpose/Aim	Design	Sample	Measures	Analysis	Relevant Results	Strengths/Limitations
				researchers) Cronbach's alpha of the Scale of Social Interest=.95; Cronbach's alpha of the Happiness of Being Scale-Revised=.90.	(Independent variables: demographic variables, type of SB, disability status, SES, number of friends, professional support, self-acceptance, feasibility of future goals, Scale of Social Interest ; dependent variables: scale score of Happiness of Being Scale-Revised and its subscales)	with SB ( $\beta=.32$ ), feasibility of future goals ( $\beta=.17$ ), feelings of belonging to society and trust ( $\beta=.50$ ), and sense of self-affirmation ( $\beta=.28$ ) explained 70% of the variance in happiness subscale score, (3) Number of friends with SB ( $\beta=.39$ ) and sense of self-affirmation ( $\beta=.60$ ) explained 54% of the variance in life-satisfaction subscale score, and (4) Number of friends with SB ( $\beta=.48$ ), feasibility of future goals ( $\beta=.26$ ), and feelings of belonging to society and trust ( $\beta=.28$ ) explained 27% of the variance in optimism subscale score.	<b>Limitations:</b> Small sample size.
Fujita, Y., & Sueda, K. 2009 <sup>b</sup>	To examine the relationships among happiness, inferiority feeling and social interest in young adults with congenital and acquired disabilities.	Quantitative, Descriptive, cross-sectional	30 young adults with SB (age 18-35 yrs), 18 young adults with spinal cord injury (age 19-39 yrs), and 40 healthy controls (age 19-29).	Kyodotai kankaku-shakudo [Social Interest Scale], Rettokan shakudo [Inferiority Feeling Scale], Kaitei Happiness of Being shakudo [Happiness of Being Scale-Revised]	Factor analysis, ANOVA, t-test, multiple regression (Dependent variables: subscales of the Happiness of Being Scale-Revised=happiness, life satisfaction, and optimism; independent variables: feasibility of future goal, feeling of self-affirmation, basic trust, feeling of contribution to others, inferiority feeling coming from lack of confidence, and inferiority feeling coming from others' cognition)	<b>Between-groups comparisons:</b> Basic trust subscale of "happiness" was significantly higher in young adults with spinal cord injury ( $p<.01$ ). Feeling of contribution to others subscale was significantly higher among healthy controls and those with SB than in those with SCI ( $p<.01$ ). Optimism subscale was significantly higher among those with SB and SCI than healthy controls ( $p<.01$ ). <b>Among young adults with SB:</b> There was no significant association between happiness scores and age/gender/employment. Results of multiple regression analyses showed that: (1) Basic trust had significant association with happiness ( $p<.01$ ), and the regression model was statistically significant ( $R^2=.54$ ). (2) Basic trust and feeling of contribution to others had significant associations with life satisfaction and the model was statistically significant ( $R^2=.90$ ). (3) Basic trust and inferiority feeling coming from lack of confidence had significant associations with optimism ( $p<.01$ and $.05$ , respectively) and the model was statistically significant ( $p<.01$ , $R^2=.64$ ). Feasibility of future goal was not significantly associated with any of the happiness subscales.	<b>Strengths:</b> Focused on young adulthood. Use of standardized measuring instruments. Use of inferential statistical tests. The authors indicate that the instruments are standardized with college students without disability. Further, the authors performed factor analyses to evaluate validity with individuals with disability. <b>Limitations:</b> Small sample size with convenience sampling. No evidence of internal reliability or stability of instruments provided.

Author(s)	Purpose/Aim	Design	Sample	Measures	Analysis	Relevant Results	Strengths/Limitations
						There were significant negative associations between the subscales of inferiority feeling and the subscales of social interest. Feasibility of future goal was positively associated with self-affirmation ( $r=.52, p<.01$ ) and negatively associated with inferiority feeling coming from others' cognition ( $r=-.44, p<.05$ ).	
Hashizume, K., Kanamori, Y., Sugiyama, M., Tomonaga, T., Nakanishi, H., Nakanishi, H., & Terawaki, K. 2003 <sup>a</sup>	To examine the usefulness of cleansing enema in school-age children with SB.	Quantitative, Descriptive	10 patients with SB (age 7-18 yrs) and families.	Survey created by authors	Descriptive statistics	<p>Most of the participants started cleansing enema before entering or at lower grades of elementary school.</p> <p>The reasons for starting cleansing enema were being concerned about fecal incontinence at school or kindergarten, difficulty in obtaining permission to get in a swimming pool, loss of appetite or nausea due to accumulated stool, suffering from bad breath, etc.</p> <p>More than 50% of the participants practiced cleansing enema twice a week; all but one patient almost perfectly achieved fecal continence by the frequency of the procedure. No significant adverse effect was observed from the cleansing enema.</p> <p>All participants reported that they were no longer concerned about fecal incontinence at school. Other positive effect reported included better appetite, improved breath, being able to get in a swimming pool, being able to travel, and decreased urinary tract infection. All participants reported that they want to continue practicing cleansing enema.</p>	<p><b>Limitations:</b></p> <p>Small sample size. Single site study. Research question was not focused on AEA.</p>
Hori, T., Narama, M., & Yamauchi, N. 2002 <sup>c</sup>	To identify the characteristic of mother's parenting attitudes and the illness management of school aged children with spina bifida and to identify the relationship between parenting attitude and illness management.	Quantitative, Descriptive, cross-sectional	53 mothers of school-aged children (elementary and middle schools) with spina bifida	Yoiku Taido Shakudo [Parenting Attitude Scale]	Bivariate correlations, Chi-square test	<p>Mothers' scores on acceptance-focused, child-centered parenting attitude and control-focused parenting attitude decreased as child age went up.</p> <p>45% of mothers of elementary school children in lower grades entrusted their children to do CIC, and 90.9% of mothers of middle school children entrusted their children with CIC; however, the proportions of middle school students' mothers who entrusted their children with cleansing enema/enema and skin care were only 33.3%.</p>	<p><b>Limitations:</b></p> <p>Relatively small sample size. Primarily focused on early adolescence. The authors indicate that Parenting Attitude Scale has validity and reliability but no evidence is provided.</p>

Author(s)	Purpose/Aim	Design	Sample	Measures	Analysis	Relevant Results	Strengths/Limitations
Kihara, H., & Suzuki, N. 2008	To explore current life situation of the members of Spina Bifida Association of Japan (SBAJ).	Quantitative, Descriptive	1,112 members of the SBAJ Age range is not clearly stated, but seems to be 0 yr to adults.	Survey created by authors. Demographic and clinical status and life situations.	Descriptive statistics	Descriptive statistical results are provided on clinical variables, self-management, education, and employment. - The sample composed of myelomeningocele (75%), spinal lipoma (17%), and other patients (4%). - 49% of the sample were ambulatory. - 50% had hydrocephalus. 12% had epilepsy. - 2% had dyspnea. - The age at which the largest number of the subjects started doing CIC by themselves was 6. - Bowel management methods used were enema (17%), cleansing enema (14%), stool extraction (35%), and use of laxative (14%). 35% were satisfied with their bowel management; 21% were unsatisfied. - 5% had learning difficulty.	<b>Limitations:</b> Little demographic data were given. Age range of the sample was wide and not limited to AYA. 70% of responses provided by parents as proxy.
Kihara, H., & Yamaguchi, K. 2010	To examine learning ability and associated difficulties of individuals with SB.	Quantitative, Descriptive	58 members of the SBAJ (ages 1 year and older, mode=7 years) (The sample was recruited from SBAJ members who requested DVDs on CIC skills and learning difficulty.)	Survey created by authors	Descriptive statistics	There were 13 patients who were 13 yrs and older in the sample. More than half of these adolescent and young adult patients seemed to have some signs of difficulties in learning and social skills.	<b>Limitations:</b> Little demographic data were given.
Koike, H. 2007 <sup>c</sup>	To clarify processes in which recognition on bowel dysfunction is formed among individuals with SB.	Qualitative, Explorative	22 individuals with SB (ages 21-44 years; mean=33.2 years)	Semi-structured interviews	Grounded theory approach	Individuals with SB acquired bowel management methods after experiencing failures. Those bowel management issues often pose activity limitations, thus strengthen their awareness of difference from others. Adult individuals with SB tend to have a resistance to change in terms of bowel management methods; however, they also seek reliable solutions.	<b>Strengths:</b> Relatively large sample size for in-depth qualitative interviews. Analytic process was described in detail in the article.
Koike, H. 2008 <sup>c</sup>	To investigate problems and special needs people with SB have related to educational experience.	Qualitative, Explorative	8 individuals with SB who have experiences of attending both regular school and special needs school	Semi-structured interviews	Grounded theory approach	The narratives by individuals with SB revealed the following: 1) At regular schools, insufficient information on SB was provided for other healthy children, which induced school bullying. 2) At special needs schools, academic achievement tended not to be valued. 3) Individuals with SB had considerably different experiences at regular and special needs school; at regular schools they considered themselves as	<b>Strengths:</b> Analytic process was described in detail in the article.  <b>Limitations:</b> Small sample size.

Author(s)	Purpose/Aim	Design	Sample	Measures	Analysis	Relevant Results	Strengths/Limitations
						"different from others," however, at special needs schools they felt that they were "not special."	
Koike, H. 2011 <sup>c</sup>	To describe processes in which women with SB establish their self-management styles.	Qualitative, Explorative	15 women with SB (age 26-45)	Semi-structured interviews	Grounded theory approach	In the process of acquiring and establishing self-management behavior, Japanese women with spina bifida attempted to obscure their difference from others and develop their own methods of coping in order to maintain self-esteem. Categories generated include: "disability characteristics of spina bifida," "different from anyone else," "a desire to lead an ordinary life," "maintenance of self-esteem," and "adjusting themselves to the difference from others."	<b>Limitations:</b> Sample consisted only of women.
Nishi, N., & Okada, K. 2008 <sup>d</sup>	To describe how mothers' recognition changes on the disability of their children with SB based on analysis of the mothers' experiences related to children's school lives.	Qualitative, Descriptive	16 mothers of children with SB who attended regular classes of elementary and junior high schools (more than 80% of the children had hydrocephalus.)	Semi-structured interviews	Grounded theory approach	The mothers recognized urinary and bowel dysfunction of their children followed by learning disorders. The mothers experienced disappointment and sorrow when the learning disorders became obvious. Factors that affected process in which the mothers' recognition on their children's disability changed included "expansion of the children's world," "teacher's attitude," "being left holding on their own," "finding supporting communities," and "fluctuation of parental feelings."	<b>Limitations:</b> Sample consisted of mothers of school-age children and adolescents.
Noda, Y., Adachi, H., Matsuno, C., Suzuki, S., Ono, T., & Kasai, Y. 2013	To reveal experiences and their feelings of women with spina bifida who are in adolescence or have reached sexual maturation related to menstruation.	Quantitative, Descriptive and explorative	89 women with spina bifida (ages 11-39 yrs) who belong to SBA-J and their mothers.	Questionnaire developed by the authors based on their findings of a qualitative study on menstrual experiences and reproductive health of women with SB.	Descriptive statistics, t-test, Chi-square test	The concerns of women with spina bifida included issues related to bowel and bladder function impairments and difficulty in coping with menstrual care due to gait impairment. Negative feelings regarding menstruation were reported.	<b>Limitations:</b> Low response rate, sample only consisted of SBA-J members. No information on reliability and validity of the questionnaire was provided.
Oi, S., Sato, O., & Matsumoto, S. 1996 <sup>a</sup>	To examine adult outcomes of individuals with SB and to identify medico-social problems these individuals encounter.	Quantitative, Descriptive, Review of medical records	141 cases of SB (age 0.3-56 yrs). 18 cases (13.5%) over 16 yrs were analyzed in detail.	Spina Bifida Neurological Scale for neurological status.	N/A.	The issues that patients with spina bifida occulta encountered included educational, familial, and sexual difficulties. The issues that patients with spina bifida aperta encountered included depression, occasional convulsions that affected school life, and gradual decline in cognitive ability due to shunt malfunction.	<b>Limitations:</b> Age range was not limited to AEA.
Ono, T., & Inaba, Y. 2008 <sup>c</sup>	To explore factors affecting QOL of school-age children with SB.	Quantitative, Explorative, cross-sectional	78 children with SB (age 9-15 yrs), 231 healthy controls (age 9-15 yrs)	QOL questionnaire developed by Nakamura (2002). The authors revised the	Factor analysis, Mann-Whitney U-test, Chi-square test	Total QOL scores of SB children were higher than those of healthy controls ( $p < .01$ ).  No significant associations were found between QOL and urinary incontinence, fecal incontinence, or gait disturbance. Anxiety and	<b>Strengths:</b> The authors revised the instrument to make it easier for parents and children with SB to understand and complete. They also conducted FA to evaluate

Author(s)	Purpose/Aim	Design	Sample	Measures	Analysis	Relevant Results	Strengths/Limitations
				questionnaire specifically for this study. Cronbach's alpha=0.86.		distress subscale score in children who self-catheterize was lower than that in children who do not require CIC ( $p<.01$ ). The factor analysis yielded following 5 factors: (1) satisfaction with friendship, (2) anxiety and concerns, (3) satisfaction with home and family, (4) satisfaction with teachers, and (5) satisfaction with general health status.	validity and Cronback alpha to evaluate internal reliability. Limitations: Primarily focused on early adolescence.  <b>Limitations:</b> Some statistical values for Mann-Whitney U test and Chi-square test were not reported.
Suzuki, S., Matsuno, C., Noda, Y., & Adachi, H. 2014 <sup>c</sup>	To reveal how women with spina bifida and their parents think about sexual issues.	Quantitative, Descriptive, correlational	113 women with spina bifida (24 elementary school children, 18 junior high school students, 15 high school students, 56 working adults under age of 40 /college students) and 121 parents (3 fathers, 118 mothers)	Questionnaire developed by the authors based on their findings of a qualitative study on menstrual experiences and reproductive health of women with SB.	Descriptive statistics, Chi-square test	58.9% of college students/working adults had anxiety and worries related to sexual issues; the rate was significantly higher than that of elementary and junior high school students (31.0%) ( $p<.01$ ). 31.5% of the parents of elementary and junior high school children, 33.3% of the parents of high school children, and 26.9% of the parents of college students/working adults showed anxiety and worries related to sexual issues. 33.3% of high school students and 41.1% of college students/working adults had someone to turn to when they need advice on sexual issues. 60.0% of high school students and 51.8% of college students/working adults have thought about romantic relationships, marriage, childbirth, and child-rearing. 26.9% of parents of college students/working adults had concerns related to sexual issues while only 10.7% of the women with SB did. The proportion of the parents who had concerns was significantly higher than that of the women with SB ( $p<.05$ ).	<b>Limitations:</b> Low response rate (26.0% for women, 27.8% for parents), sample only consisted of SBAJ members. No information on reliability and validity was provided for questionnaire.
Take, H. 2004 <sup>a</sup>	To explore current situation regarding impaired excretion in children with SB in the author's institution.	Quantitative, Explorative	32 children with SB (age 6-15 yrs)	Questionnaire developed by the author to examine bowel function and methods of bowel management.	Descriptive statistics	81% of the participants reported the symptom of having firm stools. 44% experienced incontinence every day; approximately 75% of the participants experienced frequent incontinence. 44% of the participants practiced manual removal of stools. 25% were able to evacuate the bowels on their own. In most cases parents provided their children with care in bowel management; no participants practiced their bowel management by themselves.	<b>Limitations:</b> Relatively small sample size. Not focused on AEA. Single site study. Survey with no validity and reliability
Yoshida, H., & Iwaya, T. 2000 <sup>a</sup>	To examine quality of life in individuals with spina bifida and	Quantitative, correlational	22 individuals with spinal lipoma (ages 13-42 years)	SF-36	Descriptive statistics, Spearman's rank-order correlation	The physical functioning subscale score of individuals with spinal lipoma was significantly associated with their paralysis states ( $r=0.69$ ). Subscale scores of the sample were significantly	<b>Strengths:</b> Use of standardized instrument (SF-36) to measure QOL.

Author(s)	Purpose/Aim	Design	Sample	Measures	Analysis	Relevant Results	Strengths/Limitations
	relationship between QOL and paralysis state.					lower than normative scores of Japanese healthy controls in role-physical and role-emotional domains. (Subscale scores of 4 women with myelomeningocele were not significantly different from normative scores.)	<b>Limitations:</b> Relatively small sample size. Not peer-reviewed
<p>All significant results reported were at <math>p=.05</math> unless specifically noted.  Discipline: a) medicine, b) psychology, c) nursing, d) education  All the studies were rated at level 6 in the rating system developed by Melnyk and Fineout-Overholt (2010).  *English abstract available.</p>							

### **Summary of the Two Literature Reviews**

Two literature reviews were conducted, a review of manuscripts on AEA with SB published in English and one of manuscripts published in Japanese journals. In both manuscripts, AEA with SB were faced with social and psychological challenges during transition to adulthood. In both reviews, it was also found that the AEA and their families had unmet service needs in the domains of healthcare and social adaptation. Based on these findings, a qualitative study of AEA with SB in Japan was undertaken to better understand experiences of the AEA during transition to adulthood.

## **CHAPTER THREE**

### **Research Methods**

#### **Purposes/Research Questions**

The purpose of this study was to describe the experiences of transition into adulthood for emerging adults with spina bifida (SB) in Japan. The research question for the study was “What are the experiences of transition into adulthood for emerging adults with SB in Japan?”

#### **Methods**

This study employed a qualitative descriptive design. The qualitative descriptive method was chosen for this study to explore the experiences of emerging adults with SB living in Japan during transition and meanings attached to their experiences in detail. Data were collected by conducting individual face-to-face interviews with the emerging adults with SB that focused on their experiences during transition. The Spina Bifida Association of Japan (SBAJ) offered the researcher opportunities for participant recruitment. The SBAJ is the nationwide organization of individuals with SB and their families. It has approximately 1,070 members and its activities include publication of newsletters, telephone consultation services, negotiation with government, and collaboration with study groups (Spina Bifida Association of Japan, 2014). The SBAJ currently has its local chapters in 32 prefectures (SBAJ, 2014).

#### **Participants and Sampling Procedures**

Study participants included eight emerging adults with SB who were 20 to 29 years of age and living in Japan. This age range was chosen because, as stated in Chapter 1, Arnett (2015) defines an emerging adult as a young person in this age range, during which he or she experiences more independence than adolescence and at the same time more instability and

uncertainty than adulthood. The upper age (29) was included because the age when young people typically achieve developmental milestones of adulthood, such as marriage and entry into parenthood, have been constantly rising in Japan (Minister of Health, Labour and Welfare, 2016); therefore, an individual at the age of 29 years may be considered immature as an adult in contemporary Japanese society. Age 20 was identified as the lower age range for EA as it is the age of consent for research in Japan.

In this study, purposeful sampling (Ritchie, Lewis, & Elam, 2003; Sandelowski, 1995) was employed as the sampling method to achieve the following sampling goals: (a) relevance of the sample to the topic explored in the study, namely, the experiences during transition to adulthood for Japanese emerging adults with SB, (b) homogeneity of the sample in terms of the presence of the condition (SB) and disability that similarly affects transition process for the participants, and (c) variation in the participants' experiences, such as trajectories of their transition, factors affecting their experiences, and challenges and/or positive aspects perceived during transition. It was anticipated that the sample size of ten would be adequate to describe the experiences during transition for a subgroup of emerging adults with SB in Japan that have both similarities and differences in the ways in which their daily activities are affected by their condition and disability (Patton, 2002; Sandelowski, 1995).

The inclusion criteria were: (a) a diagnosis of myelomeningocele, (b) ages between 20 and 29 years, (c) having no other major health problem(s) unrelated to spina bifida, (d) able to give consent for participation, and (e) Japanese speaking. The exclusion criteria were: Spina bifida diagnoses other than myelomeningocele, outside age range, and having moderate or severe intellectual disabilities. Variety in age, gender, living situation, and educational or employment

status was sought when recruiting the participants.

The recruitment of the participants was conducted through the SBAJ using the purposeful sampling method. Before the initiation of the recruitment, approval was obtained from the Institutional Review Boards of the University of Wisconsin-Milwaukee and of the Kobe University in Japan. After obtaining IRB approval, the researcher sent a written request for cooperation in the study to the president of the SBAJ and obtained the president's written consent. Advertisement materials with the president's cover letter were sent to potential participants. If the potential participant who was interested in the study contacted the researcher, the researcher made a telephone or email contact with them to explain the research purpose and procedure and proceed to recruitment. Interested potential participants were screened using the inclusion criteria, and those who met inclusion and retained interest had the research procedure explained in detail. Informed consent was obtained from willing participants.

### **Ethical Considerations**

Approval was obtained from the Institutional Review Boards of the University of Wisconsin-Milwaukee and of the Kobe University in Japan (See Appendices D and E). The research purpose and procedures were fully explained to prospective participants using the consent form and the researcher fully answered any questions from prospective participants. The voluntary nature of the study was emphasized and participants were assured before the interview that they could end the interviews at any point or omit a response to any questions. It was also explained to the participants that if the participant was distressed or depressed, or experiences any physical health problems during the interview, the researcher would terminate the interview, if needed, and assist the participant so that the psychological or physical health

problems would be addressed appropriately. In the current study, special attention was paid during the data collection so that the health and comfort of the participants, who had physical disability, was not compromised (Polit & Beck, 2008). Non-judgmental, empathetic listening was employed by the researcher using the semi-structured interview guide.

Also, measures to assure confidentiality were discussed with the participants before the interviews began. Recorded data and transcripts were protected with a password and were not stored with identifying information. The researcher's Major Professor and professors on the dissertation committee had access to transcripts during the process of data analysis. In addition, a Japanese researcher who understood English and had experiences in qualitative research in sociology confirmed accuracy of the translation of transcripts. The names of the participants were replaced with pseudonyms. In addition, contextual details that may lead to identification of the participants were slightly altered in the dissertation. Use of quotes in any written report was discussed with the participants and agreed upon before the interviews.

### **Data Collection Procedures**

Individual in-depth semi-structured interviews were chosen as the data collection method in this study. Once the study was described to the potential participant, a time for review of the consent form and the first interview was arranged. The interviews were conducted after obtaining written consent and both were conducted in Japanese. Consent for follow-up interviews and member checking sessions was also be obtained in advance.

Individual in-depth interviews with Japanese emerging adults with SB were conducted by telephone or Skype in their primary language (Japanese). Scheduling the interview with a method of the participant's choice was chosen to enhance the participants' comfort in telling their

experiences.

The Demographic and Clinical Information Form (DCIF) (see Appendix A) was used to gather characteristics of the sample. The demographic data included age, gender, the participant's living situation, educational type and level, and employment status. Clinical data included diagnosis, history of hydrocephalus, hospitalization history, continence status, and ambulation status. The researcher asked participants the questions of the DCIF before the interview because the discussion on those questions helped set the stage for in-depth interview about the participants' experiences during transition.

During the interviews, the researcher asked the participants to tell their experiences related to transition into adulthood using a brief interview guide developed based on the Theory of Emerging Adulthood (Arnett, 2000), the Ecological Model of Secondary Conditions and Adaptation (Sawin, Buran, Brei, & Fastenau, 2003), and the Individual and Family Self-Management Theory (Ryan, 2009; Ryan & Sawin, 2009; Ryan & Sawin, 2014; UWM Self-Management Science Center Working Group, 2011; UWM Self-Management Science Center, 2017) (See Appendix B). The interview guide consisting of open-ended questions was used to elicit the participants' responses. As compensation for research participation, gift cards that had a value of the Japanese equivalent of approximately \$20 USD each were provided to the participants at the interviews.

The interviews were recorded on a compact digital recorder, transcribed first in Japanese by the researcher. The first four interviews were then translated into English verbatim and double checked for the accuracy of translation by a Japanese researcher who majored in English translation studies in graduate school and had experiences of qualitative research in sociology.

The English transcripts were then coded under the supervision of two members of her dissertation committee who had extensive experience in qualitative research and in research on self-management and transition of individuals with SB. The coding of the interviews was double checked by these two dissertation committee members. After verifying the accuracy of the analytic process of these four interviews, the researcher proceeded to coding in English directly from Japanese transcripts.

## **Data Analysis**

**Qualitative data analysis method.** In this study, the goal of the data analysis was to identify themes that describe experiences of Japanese emerging adults with SB during transition to adulthood and the meanings the emerging adults attach to their experiences. Variations in the emerging adults' experiences were also be explored.

The qualitative data analysis for this study consisted of several stages to move to higher levels of abstraction and identify themes (Miles & Huberman, 1994, Ritchie, Spencer & O'Connor, 2003). The process consisted of two phases: (a) data management and (b) data analysis. In the data management phase, first, after reviewing the transcripts to become familiar with the interview data, initial coding was done by summarizing the meaning of each segment that relates to the research question and creating a short label. Relevant experiences were identified by reference to the guiding theories, namely, the Theory of Emerging Adulthood (Arnett, 2000), the Ecological Model of Secondary Conditions and Adaptation (Sawin, Buran, Brei, & Fastenau, 2003), and the Individual and Family Self-Management Theory (Ryan, 2009; Ryan & Sawin, 2009; Ryan & Sawin, 2014; UWM Self-Management Science Center Working Group, 2011; UWM Self-Management Science Center, 2017).

The codes were developed inductively, flexibly using Lofland's coding scheme as a guide, in which codes were categorized into the following six groups: acts, activities, meanings, participation, relationships, and settings (Miles & Huberman, 1994). Second, the initial codes were revised as data collection continued and the existing codes did not fit new data. Coding and recoding continued until all data were classified using the set of codes and regularities were found in the data (Miles & Huberman, 1994). The codes were then defined operationally (Miles & Huberman, 1994). The definitions of codes were developed and revised as the data analysis proceeded. Analytical notes were written during the data analysis to document analytical ideas and enhance the processes of "moving from empirical data to a conceptual level" (Miles & Huberman, 1994, p.74). Techniques, including clustering and making comparisons (Miles & Huberman, 1994) were used to facilitate the analytic process.

The analytic process was iterative, i.e., it repeatedly moved forwards and backwards between the levels of abstraction. Through this process, main themes that captured the experiences of Japanese emerging adults during transition were identified and descriptions of the themes were developed. In the written report of the findings, excerpts from the interviews were displayed to illustrate the themes and their range and variation.

**Techniques for establishing rigor.** In qualitative research, methodological rigor is assessed using criteria that are different from those in quantitative research. In this study, the following techniques were used to meet the standards of rigor for qualitative research (Lincoln & Guba, 1985): techniques to enhance credibility, techniques to enhance transferability, and techniques to enhance dependability and confirmability.

**Techniques to enhance credibility.** To minimize the influence of environments on the

interview data and reduce the burden on participants, the researcher offered options for interview methods (i.e., in-person, telephone or Skype interview). As a result, participants chose either telephone or Skype interviews and therefore were able to participate in the interviews at their home or locations familiar to them. Timing and environment of the interview were documented to help assess the influence on the data. In addition, the primary researcher analyzed the data with constant supervision of the faculty who had extensive experience in qualitative research and in research on self-management and transition of individuals with SB. Specifically, the interview context, including the timing of when and the environment where participants were interviewed was documented when that context may have affected the quality of data. Also documented were the social, cultural, and health care contexts that may have affected participants' experience. These contextual matters were discussed with the member-checking faculty to enhance understanding of interviews' meaning.

**Techniques to enhance transferability.** The current study, which employed social constructivism as guiding analytic philosophy and used descriptive qualitative approach, generated knowledge dependent on the context of Japanese emerging adults with SB. Therefore, generalizability to broader context is not a suitable standard of rigor for the study. Rather, rich description of the reality from the participants' perspective in a local context was prioritized (Guba & Lincoln, 1998). Nevertheless, the researcher explicated the contexts where the general patterns and themes were identified so that the reader is able to judge its transferability to a new context. The researcher herself was born and raised in Japan and educated as a nurse with clinical experience in the population of children with SB. Thus, the analysis was conducted from the perspective of a member of the community and society.

**Techniques to enhance dependability and confirmability.** Documentation on the context where the research was conducted, methodological decisions, analytic decisions, and personal responses to research elements was done to enhance dependability and confirmability (Rodgers & Cowles, 1993). These elements of the research process were constantly discussed with the faculty in the dissertation committee who had expertise in qualitative research and in research on self-management and transition of individuals with SB.

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## CHAPTER FOUR

Transition to Adulthood: Experience of Youth with Spina Bifida in Japan

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## Abstract

**Background:** Emerging adults with spina bifida have complex health care and developmental care needs. As increasing proportion of children with spina bifida reach adulthood due to improvement in neurosurgical care, transition to adulthood for this population has been receiving increasing attention. However, the experiences during transition for emerging adults with spina bifida in Japan remains unknown. The purpose of this study was to describe the experiences of transition to adulthood for emerging adults with spina bifida in Japan.

**Methods:** Qualitative descriptive design was used to explore experiences of emerging adults with spina bifida. Eight emerging adults aged between 20 and 29 years participated in semi-structured interviews.

**Results:** Six core themes were identified: (a) struggling, (b) finding the meaning of “being an adult”, (c) learning to implement self-management, (d) health care, (e) processes and challenges in establishing and retaining relationship with friends, and (f) broadening scope of experiences.

**Conclusions:** This study contributed to identification of both challenges and positive aspects in experiences of emerging adults with spina bifida in Japan. Ever present were participants’ experiences of struggling during transition to adulthood, especially when trying to find a job, establishing relationship with friends, and making transition to adult health care. Participants shared their unique perspectives on the meaning of “being an adult” as a person with spina bifida in Japan. Programs assisting emerging adults with spina bifida in achieving milestones for adulthood and transitioning to adult health care in Japanese social context are needed.

*Key words:* spina bifida, emerging adult, transition to adulthood, self-management, transition to adult health care

## Introduction

Spina bifida (SB) is a congenital chronic condition that affects functions of multiple body systems, including orthopedic, urologic, gastrointestinal, neurological, and cognitive. The prevalence of SB in Japan was 5.18 cases per 10,000 births in 2012 (International Clearinghouse for Birth Defects Surveillance and Research, 2014). While the prevalence of SB is generally declining or stable in European and North American countries, an increase has been observed in Japan (International Clearinghouse for Birth Defects Surveillance and Research, 2014).

An increasing proportion of children with SB grow into adulthood due to advances in neurosurgical management (Bowman, McLone, Grant, Tomita, & Ito, 2001). As a result, issues related to their transition to adulthood received more attention. In the context of developmental and health care needs of individuals with SB, two components of transition to adulthood need to be considered: transition to adult life and transition from child-centered to adult-centered health care. The Guidelines for the Care of People with SB (Spina Bifida Association, 2018) explicitly addresses both achievement of developmental milestones including self-management behaviors and health care transition for this population.

Theory of emerging adulthood provides a theoretical basis for the study. Emerging adulthood is a developmental stage that typically ranges from 18 to 29 years of age (Arnett, 2015). According to Arnett (2015), young people in developed countries nowadays enter adult life (i.e., a stable job, marriage, and parenthood) later due to changes in society. Emerging adults seek more independence from their family; however, they have not yet achieved firm commitment to adult life (Arnett, 2015). While this theory was originally developed in the United States, the later move to adulthood likely exists in Japan since the tendency of longer education and delayed marriage and parenthood in young people is also observed.

Emerging adults with SB are at risk of having challenges during transition to adulthood. Studies on psychosocial issues in adolescents and emerging adults with SB showed that they were faced with various challenges, including self-management demands, social isolation, stigma, and psychological distress (Kinavey, 2007; Ridosh, Braun, Roux, Bellin, & Sawin, 2011; Sawin, Bellin, Roux, Buran, & Brei, 2009). On the other hand, experiences that could be protective of their healthy transition were also described, including development of inner strength (Ridosh et al., 2011), social and professional support (Lindsay, McPherson, & Maxwell, 2017).

Existing research literature that specifically focused on the experiences of emerging adults with SB in Japan is limited. The process in which adolescents and emerging adults in Japan learned self-management behaviors (i.e., clean intermittent catheterization, bowel programs, and skin care) was described in several studies (Hori, Narama, & Yamauchi, 2002; Kihara & Suzuki, 2008; Koike, 2007, 2011; Take, 2004). Only two studies addressed psychological and social process in which adolescents and emerging adults with SB learned to practice self-management behaviors qualitatively (Koike, 2007, 2011). Health care needs of these adolescents and emerging adults as well as their families were also explored. Individuals with myelomeningocele, the most severe form of SB, had concerns about self-management, cognitive status, education, employment, and mental health (Date & Ito, 2003; Oi et al, 1996). One study addressed transition to adult health care for individuals with SB, revealing that among those who had received care at a facility specializing in pediatric health care in childhood, 40% continued to be seen in the same health care facility at age 18 years and older (Domae, Date, & Sakuda, 2014). Development of a multidisciplinary SB program was a focus of interest for

some clinicians. The purposes for the patients' visit of the multidisciplinary SB clinic developed included transition to adult life and to adult health care (Date, Ito, Numata, & Matsuoka, 2005)

Several studies addressed mental health and social adaptation of adolescents and emerging adults with SB in Japan. One study showed that subjective well-being was significantly lower in emerging adults with SB than those without the condition (Fujita, 2010). In another qualitative study, individuals with SB reported their sense of difference from others at regular school (Koike, 2008). The study showed difference in those individuals' social adaptation experiences between special education settings and regular schools (Koike, 2008).

These studies demonstrated that adolescents and emerging adults with SB in Japan were faced with various psychological, social, and health challenges (Date & Ito, 2003; Domae et al., 2014; Oi et al, 1996; Koike, 2008, 2011). However, these studies often used samples with age ranges wider than that of emerging adulthood. Further, few studies described the experiences of emerging adults with SB during transition to adulthood and what those experiences meant to them. Thus, qualitative research on the experiences of the emerging adult during transition to adulthood is needed.

The purpose of this qualitative study was to describe the experiences of transition to adulthood for emerging adults with SB in Japan. The research question for the study was "What are the experiences of transition to adulthood for emerging adults with SB in Japan?"

## **Methods**

### **Design**

This study used a qualitative descriptive design. The qualitative descriptive method

was chosen for this study to explore the experiences of emerging adults with SB living in Japan during transition and meanings attached to their experiences in detail. Data were collected by conducting individual telephone or Skype interviews with the emerging adults with SB that focused on their experiences during transition.

### **Participants and Sampling Procedures**

Purposeful sampling (Ritchie, Lewis, & Elam, 2003; Sandelowski, 1995) was employed. The inclusion criteria were: (a) a diagnosis of myelomeningocele (Spina Bifida Association, 2021), (b) ages between 20 and 29 years, (c) having no other major health problem(s) unrelated to SB, (d) able to give consent for participation, and (e) Japanese speaking. The age range was derived from Arnett's (2015) definition of an emerging adult as a young person in this age range, during which he or she experiences more independence than adolescence and at the same time more instability and uncertainty than adulthood. The exclusion criteria were: Spina bifida diagnoses other than myelomeningocele, outside age range, and having moderate or severe intellectual disabilities.

The recruitment was conducted through the Spina Bifida Association of Japan (SBAJ). At the time of the study the SBAJ had 32 chapters across Japan, 1,070 members and a number of resources including newsletters, telephone consultation services, negotiation with government, and collaboration with study groups (Spina Bifida Association of Japan, 2018). After obtaining human subjects approval, the approval for recruitment was granted by SBAJ. The president of the SBAJ sent advertisement materials with the president's cover letter to potential participants using their membership list. Potential participants contacted the researcher, who explained the research purpose and procedure and proceeded to recruitment. Interested potential participant

were screened using the inclusion criteria. The consent form and procedure were explained in detail to the potential participants before any data collection began. In this study, eleven individuals contacted the researcher. Two individuals could not be reached after they expressed an interest in the study and one had an unexpected hospitalization. Consequently, eight individuals participated in the interviews. The data were collected from April to August 2019.

### **Ethical Considerations**

Approval was obtained from the Institutional Review Boards of the University of Wisconsin-Milwaukee and of the Kobe University in Japan (see Appendices D and E). During consent, participants were informed that if they were distressed or depressed, or experienced any physical health problems during the interview, the researcher would terminate the interview, if needed, and provide the participants with referral to appropriate resources. Before finalizing consent, the researcher administered a screening form to assess the individual's understanding of the purpose of the study and provide consent. In this study, the IRB approved the option to obtain verbal consent from all the participants because the interviews were conducted by telephone or Skype. The consent was documented in the transcript. To assure confidentiality, recorded data and transcripts were password protected and stored in a de-identified manner.

### **Data Collection Procedures**

Data were collected using individual in-depth semi-structured interviews. The telephone ( $n = 5$ ) or Skype ( $n = 3$ ) interviews were conducted in their primary language (Japanese). A Demographic and Clinical Information Form (DCIF) (see Appendix A) was used to gather characteristics of the sample. The demographic data included age, gender, the participant's living situation, educational type and level, marital status, and employment status.

Clinical data included diagnosis, history of hydrocephalus, history of hospitalization and surgery, bladder and bowel programs, and ambulation status.

During the in-depth interviews, the participants were encouraged to share their experiences. An Interview Guide with open-ended questions was developed based on three related theoretical approaches to the transition period: (a) Theory of Emerging Adulthood (Arnett, 2000, 2015), (b) Ecological Model of Secondary Conditions and Adaptation (Sawin, Buran, Brei, & Fastenau, 2003), and (c) Individual and Family Self-Management Theory (Ryan, 2009; Ryan & Sawin, 2009; Ryan & Sawin, 2014; UWM Self-Management Science Center, 2017) (See Appendix B). The interviews were recorded, transcribed verbatim by the researcher first in Japanese, and then translated into English. The translations were double checked by a Japanese researcher experienced in English translation studies and qualitative research. Participants received a Thank You gift card with the Japanese equivalent of approximately \$20 USD after the interviews. All the interviews were conducted before the COVID-19 pandemic.

### **Data Analysis**

Data analysis for this study consisted of several stages to move to higher levels of abstraction and identify themes (Miles & Huberman, 1994, Ritchie, Spencer & O'Connor, 2003). After reviewing the transcripts to become familiar with the interview data, initial coding was conducted by summarizing the meaning of each segment that related to the research question and creating a short label.

The initial codes were revised as data collection continued and the existing codes did not fit new data. Coding and recoding were continued until all data were classified using the set of codes and patterns were found in the data (Miles & Huberman, 1994). The codes were then

defined operationally (Miles & Huberman, 1994). Analytical notes were written during the data analysis to document analytical ideas and enhance the processes of “moving from empirical data to a conceptual level” (Miles & Huberman, 1994, p.74). Techniques, such as clustering and making comparisons (Miles & Huberman, 1994) facilitated the analytic process. Through this process, main themes that captured the experiences of Japanese emerging adults during transition as well as sub-themes were identified, and descriptions of the themes were developed.

Several techniques were used to enhance the standards of rigor for qualitative research including credibility, transferability, dependability, and confirmability (Lincoln & Guba, 1985). Credibility was addressed by documenting and assessing the influence of timing and environment of the interview. In addition, data were analyzed with constant supervision of two researchers in nursing who had extensive experience in qualitative research and in research on self-management and transition of individuals with SB. Specifically, the interview context, including the timing of when and the environment where participants were interviewed was documented when that context may have affected the quality of data. Also documented were the social, cultural, and health care contexts that may have affected participants’ experience. These contextual matters were discussed with the member-checking faculty to enhance understanding of interviews’ meaning. Transferability was addressed by explicating the contexts where the general patterns and themes were identified. Documentation on the context where the research was conducted, methodological decisions, analytic decisions, and personal responses to research elements was done to enhance dependability and confirmability (Rodgers & Cowles, 1993).

## **Results**

Eight participants who were 20 to 29 years old were interviewed. (See Table 4.1). All participants were single and living with one or both of their parents. One participant resided in a dormitory on weekdays. Two participants received special education during junior high school and high school. One participant was attending college and two were in vocational training programs. One student also worked part-time and only three worked full-time. Sixty-two percent of participants had a shunt, 75% used a wheelchair for mobility, 87.5% performed intermittent catheterization to manage the neurogenic bladder and 50 % used a cleansing enema for their bowel program. Five participants participated in the interviews by telephone and three by Skype. The interviews lasted for 70 to 160 minutes. Some demographic and clinical characteristics were slightly modified for confidentiality due to the rarity of SB in Japan.

Six core themes were identified from the interview data: (a) struggling, (b) finding the meaning of “being an adult”, (c) learning to implement self-management, (d) health care, (e) processes and challenges in establishing and retaining relationship with friends, and (f) broadening scope of experiences. Several concepts were present in more than one theme. The concept of self-management was present in the themes of “struggling,” “finding the meaning of being an adult” and “health care.” Also, the concept of struggling was present in the theme of “processes and challenges in establishing and retaining friends.” The six core themes had 16 subthemes distributed across the themes which will be discussed below (See Table 4.2 for themes, sub-themes and exemplar quotes).

### **Struggling**

This theme captured the participant’s experiences of struggling in several different areas of transition to adulthood described in two sub-themes: (a) *struggling to find a job and work* and

(b) *struggling with relationship with family.*

**Struggling to find a job and work.** Participants struggled to find a job and to work. Their struggles with finding a job were caused by lack of understanding or accommodation of their special health needs at workplace and lack of legitimate appraisal of their ability. Some participants also implied that they felt the employers' reluctance to hire individuals with disabilities during the process of searching for a job. Only two participants reported that they received professional support when transitioning to employment. One participant felt caught between different opinions around job-searching expressed by the people around her.

*Well, I guess I was having the most difficult time at that time. Well, at home I was told, "You can attend the vocational rehabilitation and take it easy," however, at school, well, you know, there were other students, too, so it was what they were saying to those students, but they emphasized "brand value of new graduates" saying, "The best thing to do is to join the company as a new graduate." and I heard that all the time. ... So, I was like, "So, which one should I trust?" and I guess I was having the most difficult time at that time. (344-349, 354-355, 03)*

In addition, several participants worked jobs while dealing with challenges, including an inaccessible work environment and internal culture of unwritten rules at the workplace that did not fit the participants. Some participants also reported lack of understanding and accommodation of their special health needs at the workplace and described their challenges in protecting their own health while working. They had difficulties with preventing pressure sores or securing time to catheterize regularly during work. One participant explained that it was difficult for him to communicate with his co-workers about his health care needs.

Further, another participant reported that his ability was not appraised appropriately by the employer and that he could not build his career due to such inappropriate appraisal to his ability and lack of the employer's understanding.

*Nonetheless, like I said earlier, my co-workers in their 50's always rely on me when it comes to computer skills. However, I get paid less than anyone else because of my employment contract, I'm a bit dissatisfied with that, though. (684-688, 08)*

**Struggling with relationship with family.** Participants had ambivalent emotions toward the relationship with their family. They were thankful to their parents for the care and support they had received, both material and psychological, since childhood.

*Anyway, my mother gave me the opportunity to join the [band], and by joining the [band] ..., I think that the [band] was the biggest trigger leading to my personal growth. (138-141, 03)*

However, some participants described their struggles with the relationship with their family. They experienced disagreement with their family and/or felt that it required effort to live with their family.

*I have to pay attention to the time because I live with my family, and I have to cooperate [with other family members] because I am not living alone. (645-651, 02)*

### **Finding the Meaning of Being an Adult**

Participants actively engaged in finding the meaning of "being an adult." The two sub-themes included: (a) *trying to achieve what the common definition of "being and adult" means* and (b) *exploring the meaning of "being an adult" for a person with SB*. Participants explored these two different mental images of an adult simultaneously.

**Trying to achieve what the common definition of "being an adult" means.** The participants shared that the goals they set reflected what individuals might typically aim for during transition to adulthood. Such goals included becoming mature, learning to take care of oneself, becoming able to take responsibility, and living on one's own.

*I'm aiming to live on my own in the future. Although I would say I am satisfied with my current environment, my goal is to live on my own. (598-599, 04)*

**Exploring the meaning of "being an adult" for a person with SB.** Participants also explored what "being an adult" meant uniquely for a person with SB. Three participants described what they felt about their actual life situations when compared to the goals they had set or to the goals people commonly set during transition to adulthood. One participant shared his pain due to his inability to achieve the goals people might typically achieve when they reached adulthood.

*I feel that it would be painful if someone says that I'm not a fully grown adult because I'm living with my parents and can't earn enough money to live on my own. (37-39, 08)*

Another participant reported that he did not have an opportunity to accomplish a goal due to the influence of his parents' thoughts on his life.

*I had a chance to [live on my own] once when I was in college, but I was told not to leave home by my parents. So, I've never really left home. And, I'm out of my parents' home now just because there is a dorm at my school, and it seems that my parents hope that I live in their home for life. So, I've never had an opportunity to feel strongly that I've become an adult, like I've grown up. (27-32, 01)*

The participants perceived the differences between what was commonly expected as an

adult and their actual life situations, thus exploring what “being an adult” meant uniquely for them. One participant shared his perspective on what it meant to become an adult for a person with SB.

*When it comes to people with this condition, if you consider those who can do everything by themselves as fully grown adults, I feel that that will mean I haven't become able to do anything yet. Well, I think that one could be considered a fully grown adult as a person with SB if he can go out for a certain distance or for a certain amount of time. (20-25, 08)*

Another participant considered learning about the condition and becoming able to implement self-management behaviors as the core elements of “becoming an adult” for a person with SB.

### **Learning to Implement Self-Management**

This theme captured the process in which the participants learned to implement self-management of their SB. The three sub-themes included: (a) *learning to implement self-management behaviors*, (b) *learning to seek help when necessary*, and (c) *preparing environment for self-management*.

**Learning to implement self-management behaviors.** This subtheme captured the process in which the participants learned to implement self-management behaviors, including bowel and bladder management, getting cleaned up when soiled, pressure sore prevention, making hospital visits, obtaining medications and medical devices (e.g., urinary catheters), and communicating with the health professional during an appointment. Participants learned to implement the self-management behaviors to maintain their health and prevent complications.

Participants described the factors that led to their learning of the self-management

behaviors. These factors included changes in their living environment, developmental changes in family relationship (e.g., the participant entering rebellious phase), and changes in family role structure.

*Well, I wonder until when..., I'm not sure whether it's embarrassing or not, but my parents did it for me until I graduated from junior high school, but I had to do the enemas in the boarding house when I was in high school, you know. Because I do it twice a week. I had to learn to do that. (391-397, 04)*

*My grandpa used to live with my family, but, I guess it's been two years already... he went into a nursing home since his [illness] was a bit severe. So, my mother got busy taking care of grandpa, and therefore, I thought that I just had to do [the self-management tasks] by myself. (533-537, 02)*

Some participants also stated that their parents' advice had an impact on their self-management behaviors. Several participants described health providers' assistance with their self-management. The participants reported their experiences of receiving health providers' advice on their self-management; however, it had differing impacts on their own behaviors.

Some participants still required family's help with performing some self-management behaviors, such as bowel management and communicating with health professionals; however, they tended to have the motivation to learn to do those tasks by themselves or already explored ways to do so.

The participants had begun to learn how to perform select self-management tasks in childhood. By the time they reached their 20's they learned not only to simply follow the procedure of the tasks but also to monitor and assess the outcomes of their self-management and

explore methods for better management. In addition, the self-management behaviors were incorporated into the participants' daily life.

*When I have lab classes, I go to the bathroom before the class begins, and stuff like that, because I can't be late or miss the lab classes. I'm always thinking about it, like I should go to the bathroom between the 1st and 2nd period, I'm thinking about those things. (266-273, 05)*

**Learning to seek help when necessary.** In addition to learning to manage their condition by themselves, the participants learned to seek help to accomplish their self-management when needed. They learned to rely on their friends and people other than family as they grew up and spent more time outside of the family unit.

*And, well, ummm, I get my diaper changed by my friends [during a trip], and I don't know if I should say "they are happy to do it," (laughs) but they just say, "We'll do it if you need help." I'm really thankful to them. (700-704, 03).*

**Preparing environment for self-management.** Participants requested understanding and accommodations at workplace when they were necessary for balancing both work and self-management. For example, some participants reported that they requested time to catheterize during work or use of a bed for pressure sore prevention. The participants also learned to use services to implement their self-management without assistance from their family. Some participants actively managed services, such as home care workers.

*At present, the [disability support] program doesn't allow me to have a care worker 24 hours a day. So, I'm talking to the social worker about transferring to the program that allows me to have the service 24 hours a day. Well, I can't actually do everything*

*on my own even if I live alone, so I think I have to do it by using the care worker's help on it. So, now, I'm thinking about it, I think I should make a transition to it little by little. (609-615, 04)*

However, not every participant was engaged in such management; there were variations in their roles in the utilization of services.

## **Health Care**

The participants described their experiences of transferring to adult health care or to health care services different from the ones they used in their childhood, including two sub-themes: (a) *transferring to adult health care* and (b) *worrying over health status and receiving health care*.

**Transferring to adult health care.** Most of the participants described their experiences of transferring to adult health care or to health care services different from the ones they used in their childhood. There were several different trajectories of transferring to adult health care. Three participants reported that they received health care from multiple specialists including neurosurgery, urology, orthopedics, and ophthalmology, at a children's hospital in their childhood. Except for one participant who continued to receive care at the children's hospital after she turned 18 years old, those participants transferred to adult health care since they were no longer eligible for care at the children's hospitals.

*Children's hospitals are for children who are 18 or younger, right? Because the government says that you are called "a child" until you are 18 years old... You know, you are a child until you are 18. So, I used to go to the children's hospital until I was 18. ... Then, one day the doctor said to me, "Now that you're already 18, you should*

*graduate from the children's hospital." So, I graduated from the children's hospital shortly after I turned 18 years old. (497-504, 04)*

Half of the participants reported that they went to multiple specialists at different health care facilities since childhood. Some participants transferred to health care services different from the ones they used in their childhood for the reasons other than transition to adult health care, such as its closeness to their houses and retirement of the physician. In addition, one participant reported that she moved from the university hospital to the nearby clinic for ophthalmology follow-up because her condition was sufficiently stable.

Three participants described their feelings about communication with the health care providers. They reported that the health care providers whom they saw in their childhood or they continued to see over the years were approachable and knew them well. They also described their smooth and frank communication with those providers on their condition and self-management.

*I've been seeing those doctors since I was little, so actually I guess, probably they think of me just as a child. It's like I go there just to spend time with them, hahaha (laughs), just to talk with them. (579-585, 03)*

Further, one participant reported that he moved from a children's hospital to adult health care, where the provider had connection with the providers at the children's hospital.

**Worrying over health status and receiving health care.** Several participants had worries over their future health status. Their worries were mainly over worsening of complications or conditions related to SB. Other participants described their concerns over receiving health care. These participants were concerned about lack of adult health providers

who had expertise in SB, possible risks in emergency situations, and financial burden of the health care.

One participant described his effort to explain both his condition and health needs to health care providers who did not specialize in SB. He found the process of receiving health care from such providers arduous.

*I have a family doctor, so it's easy in my case because I can leave the primary care to the doctor; but, when I see other doctors I have to explain my condition and what medicine I can't take due to the condition. In addition, I have to explain my whole medical history to the doctor when I'm hospitalized, and the process is so tough, even tougher than explaining my symptoms. (481-486, 08)*

### **Processes and Challenges in Establishing and Retaining Relationship with Friends**

This theme captured participants' experiences of nurturing relationships with their friends and challenges with their friendship, including five sub-themes: (a) *nurturing relationships with friends at school/in community*, (b) *nurturing friendships with peers with SB or physical disability*, (c) *spending time with friends*, (d) *struggling to be involved in romantic relationship*, and (e) *lack of peer support*.

**Nurturing relationships with friends at school/in community.** The participants described the relationships with their friends at school and in their communities. Some participants shared their positive feelings about their relationships with those friends. Continued friendships from childhood were especially valued. One participant described her strong bond with friends from high school and expressed her gratitude for the friendship that enabled her to participate in various activities, such as taking a trip.

*I really appreciate it when I enjoy myself with [my close friends from high school], and there was once a time when I said to them, "I've had a lot of great experiences," and then they just seemed comfortable saying, "Why don't you do what you think is fun?" So, I really like it. (737-740, 03)*

Some participants experienced difficulties with maintaining relationship with their friends in community. One participant shared his experience of separation from his friends in his community due to his enrolment into special needs junior high school.

*The relationship with my friends from elementary school..., they went to the neighborhood junior high school, and I went to the special needs school, so, well, my friends at elementary school, uh, I am still friends with one of them, but you know, the relationship with them got broken from that time. (72-76, 04)*

Another participant described her encounter with the biased attitude in people around her toward her condition and disability.

**Nurturing relationships with peers with SB or physical disability.** Some participants reported that they nurtured relationships with their peers with SB or with other physical disabilities. Two participants who went to special needs junior high schools and/or high schools reported that they had opportunities to nurture friendship with other students with SB or other physical disabilities at school. One participant shared his experience of growing his friendship with peers through the activities at the special needs school and the student dormitory. Another participant reported that she nurtured friendship with her peers with SB of the same age through wheelchair sports.

*It's fun to play the sport, and I could make friends there. I made friends with people in*

*the same age with the same condition. I chat with them and take a trip with them, and stuff like that. (62-65, 05)*

One participant shared her challenge with relationship with her classmates at a special needs high school. She perceived the disagreement over perspectives on career choices as a barrier to friendship.

*I went to the employment transition support program and then got a job at a regular company. Other students went to programs with different goals, and because of the difference, I felt that I could never get along with them... (343-346, 02)*

**Spending time with friends.** Participants reported that they spent time with their friends in various ways, including chatting, eating out after work, playing sports, and taking a trip.

*Probably I'm spending the most fulfilled days with my friends from high school now, those friends from the high school [band] club let me have quite an experience. I said earlier in the interview, "Of course I go on a trip with my friends" and the friends [who go on a trip with me] are two of my friends from the high school [band] club. We went on a trip during summer vacation last year. (693-698, 03)*

**Struggling to be involved in a romantic relationship.** Participants had differing feelings about being involved in a romantic relationship. Some of them had a sense of expectation and stated that they hoped to have a romantic partner. On the other hand, others expressed concerns or hesitation over getting involved in such a relationship or getting married although few participants explicitly associated the concerns with their condition/disability.

*If the person says, "Do you want to go out on a date?" but there is no accessible*

*bathroom near the place, then I won't be able to be there. I would feel sorry for not being able to fulfill the wish, but there is nothing I can do about it. (273-275, 08)*

**Lack of peer support.** Two participants sought more peer interactions and activities. The participants who attended regular schools tended to hope for more communication with peers with SB or other disabilities.

*So, when it came to the question like “what people with disabilities in the same age are like?” I had little chance to talk with people [with disability] in the same age. (105-108, 01)*

### **Broadening Scope of Experiences**

Participants described the process in which they broadened the scope of their experiences during transition to adulthood. The theme included two sub-themes: (a) *creating opportunities by learning to get around on one's own* and (b) *barriers to broadening scope of experiences*.

**Creating opportunities by learning to get around on one's own.** Many participants became able to get around by public transportation, by car, or using wheelchair, thereby broadening the scope of their experiences. By learning to get around without their family's assistance, they had more opportunities to enjoy spending time with friends. Also, one participant reported that he became able to enjoy spending time alone. This impact of being able to get around on one's own was not limited to the broadened sphere of activities; it also influenced various aspects of life, including encounters with people and a way of thinking.

*By going to school by train, I met many people, and I encountered various emotions, and in myself I think I probably wouldn't have thought like this if I hadn't gone to school*

*by train. (129-131, 03)*

**Barriers to broadening scope of experiences.** Some participants shared their challenges in broadening the scope of their experiences. Lack of accessible transportation, insufficient built environment accessibility, and insufficient care worker services for individuals with disability led to the challenges. One participant reported that there were times when she gave up opportunities to go out with her friends due to lack of accessible transportation. In addition, another participant stated that he hesitated to go out with his friends because he was afraid of bothering his friends due to the limitation on his activities.

*I got my driver's license when I was a first-year college student, and so [when I was in high school] my parents gave me a ride when I went out [with my friends] and I had to tell them what time I was going home, and stuff like that. Isn't that a bit annoying in the circle of friends or students? I had to say to them, "I've got to go now."... I thought that I might end up causing them trouble... So, I just went to Karaoke with some of my close friends when I graduated from high school, but other than that, I never went out or stayed somewhere overnight with someone when I was a student. Never. (239-248, 08)*

## **Discussion**

This study addressed the gaps in research on the experiences of emerging adults with SB in Japan during transition to adulthood. The theme “Struggling” is discussed first because it permeates other themes identified in this study. While each of the other themes have been discussed in the broader literature, every theme has unique context in the lives of emerging adults in Japan.

### **Struggling**

The findings of the study suggested that emerging adults with SB in Japan had struggles with finding a job and working. This finding was consistent not only with previous research conducted in Japan (Koike, 2011) but also with existing research conducted in North America that revealed challenges in finding a job and concerns about the impact of the physical disability on work (Lindsay et al., 2017). Some participants of the study also described environmental (e.g., lack of understanding and inaccessible work environment) and personal factors (e.g., difficulty in communicating health care needs with co-workers) that led to their difficulties in working, which was also consistent with existing research (Lindsay et al., 2017). They further pointed out factors that prevented them from working and building their career, such as lack of legitimate appraisal of their ability. Employment for individuals with disabilities is limited in Japan although it has been promoted by adopting employment promotion systems, including legal employment quota at regular companies through the special subsidiary program (Matsui, 2013). This study showed that emerging adults with SB in Japan confront this issue of limited employment opportunities for individuals with disabilities.

Relationship with family was another area where participants had struggles during transition to adulthood, which was consistent with literature (Lindsay et al., 2017; Ridosh et al., 2011). Over-protectiveness and active engagement of parents in the lives of young people with SB have been found in previous research (Koike, 2011; Lindsay et al., 2017; Ridosh et al., 2011; Sawin et al., 2009). While participants did not explicitly describe their parents as over-protective, they indicated disagreement with parents over their life situations and future goals.

### **Finding the Meaning of Being an Adult**

In this study, a difference was found in the meaning of being an adult for emerging

adults with SB between western societies and Japanese society. Many studies conducted in North American and European countries assume that emerging adults with chronic conditions and/or disabilities including SB envision future goals similar to the goals emerging adults without chronic conditions and/or disabilities typically set during transition to adulthood, such as independent living. However, future goals the participants of this study envisioned were somewhat different. They did not necessarily try to achieve those typical goals; rather, they explored what “being an adult with SB” meant for themselves based on their own experiences of living with the condition.

This difference may be in part associated with Japanese social and cultural context. For emerging adults in Japan in general, becoming financially and psychologically independent as well as taking care of oneself are considered as components of being an adult (Suzuki, 1999; 2002). However, previous research showed that Japanese emerging adults attached greater importance to caring about one’s family compared to emerging adults in the U.S. (Suzuki, 2002). In addition, young people in Japan have experienced unstable employment and a decline in wages due to stagnation of the Japanese economy since mid-1990, which has led to difficulty with living on one’s own, getting married, and having children (Ministry of Health, Labour and Welfare, 2013). Thus, remaining living in parents’ home may be considered as one of the commonplace choices by emerging adults in Japan.

The Japanese social environment may lead to a delay in reaching developmental milestones for emerging adults with SB. According to the theory of emerging adulthood, emerging adults become more independent than in adolescence, and it is normative that milestones for adulthood are achieved by the age of 29 (Arnett, 2015). In this study, while most

of the participants expressed their desire for more independence, they had not achieved the higher level of independence due to financial dependence on parents, limited support for individuals with disabilities, or the message from parents that prevented them from achieving more independence. This finding implies that the social environment where dependence on family tends to be tolerated and opportunities for social participation for individuals with complex health care needs are limited has an impact on the pattern of behavior among emerging adults with SB.

As a result, emerging adults with SB in Japan may try to aim for what “being an adult” typically means and to explore the meaning of “being an adult” for a person with SB at the same time during their transitional period. The participants shared their own perspectives on transition to adulthood, in which they valued setting attainable goals and moving step by step toward them. Examples of attainable goals they described included becoming able to go out independently “to some extent,” learning about the condition and becoming able to implement self-management behaviors. Thus, the findings of this study suggest that for emerging adults with SB in Japan, it may be important to find a healthy balance between aiming for typical goals of becoming an adult and exploring what “being adult” means uniquely for individuals with SB.

### **Self-Management Behaviors**

Participants of this study shared the process in which they learned to implement behaviors needed to manage their SB. Previous research conducted in the U.S. found that peer conferences and camps provided young people with SB with opportunities to learn self-management behaviors (Ridosh et al., 2011). In this study, participants did not mention such events except for one participant who was hospitalized for the purpose of practicing self-

catheterization; instead, they described factors that led to their learning of self-management behaviors, including changes in living environment and changes in family relationship and role structure. These changes in their lives made them motivated or feel the need to perform self-management behaviors more independently.

Some participants coordinated their bowel and bladder care routines to make it fit in with their daily lives. Further, some participants stated that they monitored and assessed their self-management skills and outcomes. This finding was consistent with previous research that revealed young people's engagement in self-management activities (Sawin et al., 2009).

Preparation of environment for self-management by requesting understanding and accommodations, which had been described in previous research in a school context (Lindsay et al., 2017), was reported in the context of work by the participants of this study. The finding may imply the participants' engagement in addressing their own struggles and advocating for themselves at workplace. In addition, some participants reported that they learned to rely on the help from people outside family during the process in which they learned to implement self-management. This finding may suggest that the participants of this study, most of whom had moderate to severe physical disability, explored a feasible way to implement self-management outside the home when they broadened their range of activities.

### **Health Care**

In this study, experience of transition from pediatric to adult health care with sufficient planning and preparation was rarely reported. Instead, some participants shared their experiences of sudden transfer to adult health care and moving from hospital to hospital. While clinicians in Japan have started to develop health care transition programs for individuals with

SB (Date et al., 2005; Date et al., 2008), the number of such programs is limited. The findings of both this study and previous research (Domae et al., 2014) may reflect the limited opportunity for emerging adults with SB to receive transitional support in Japan.

This study revealed challenges emerging adults with SB were faced with in receiving adult health care, such as stressfulness in communicating with providers who were not knowledgeable about SB. Emerging adults with SB may not have encountered this type of challenge until they move from pediatric to adult health care because pediatric care facilities tend to provide comprehensive, coordinated health care services that meet their complex health care needs. Also, one participant shared concerns about receiving emergency medical care without explaining medical history to the provider. In-depth interviews may have been useful in identifying these concerns and stressfulness unique to this population shared from their own perspectives.

### **Processes and Challenges in Establishing and Retaining Relationship with Friends**

Participants of this study described how they established and retained relationships with their friends at school and in community as well as their peers with the same condition or other physical disabilities. Consistent with previous research that revealed challenges with relationship with friends both in North America and in Japan (Kinavey, 2007; Koike, 2008, 2011; Ridosh et al., 2011), the participants of this study experienced social isolation and difficulty with developing romantic relationships. On the other hand, they also reported positive feelings about spending time with friends and strengthening their bonds. Further, the findings suggested that their friendship facilitated participation in developmentally appropriate social activities, such as going out after school/work or playing wheelchair sports with peers.

One of the challenges the participants pointed out in the light of transition to adulthood was lack of peer support. Previous research has found that clinicians considered peer-mentoring as beneficial for youth and young adults with SB (Lindsay et al., 2017). Research on psychological health in emerging adults with SB in Japan (Fujita, 2014) also revealed a positive association between the number of peers with SB and sense of happiness. Some participants of this study hoped to receive more support from peers with the same condition and to draw on the experiences of the peers.

### **Limitations**

A potential limitation of this study is small sample size (n=8). In particular, romantic relationship in emerging adults with SB may not be fully understood in the current study as no participant had experience in such relationships. Another potential limitation is that the use of telephone or Skype interviews prevented the researcher from seeing nuances of communication with interview or from confirming whether each participant had sufficient privacy in the place they chose when participating in the interview. If there was a lack of privacy, this could have prevented participants from fully answering questions of personal and private nature. The use of technology to conduct interviews, might also have been a strength because this interview method allowed data to be collected from individuals who represented a wider geographical area including both urban and rural areas of Japan and was more convenient for participants who did not need to travel to an interview site.

The participants in this sample were all members of the Spina Bifida Association of Japan. As a result, the sample may consist of individuals who are more active or have more opportunities to receive support when they belong to a patient advocacy group compared to those

who do not. In this study, the sample did not include individuals with cognitive challenges. Consequently, the experiences of individuals who have cognitive or other communication difficulties may not be reflected in the findings. Also, the participants of this study had myelomeningocele and primarily consisted of individuals with physical limitations necessitating the use of a wheelchair for ambulation as well as bowel and bladder care and may not reflect the experience of those who have a milder form of SB.

### **Implication for Practice**

The participants had a shared experience of struggling when trying to find a job and working. In Japan, the main employment pathways for individuals with disabilities include (a) employment at regular companies through the legally mandated employment quota system and (b) participation in a support program providing opportunities of work or other activities for people with disabilities (Suzuki, 2016). Also, there are various programs for individuals with disabilities who seek employment, such as employment transition programs and vocational rehabilitation centers (Aizawa & Hisanaga, 2012). To find employment that fits the ability and health care needs of the emerging adult, professional support is desirable. In this study, however, few participants reported experiences of receiving professional support in a timely manner when searching for employment. Nurses and other health care providers should provide appropriate information on possible employment pathways as well as available employment support programs to the emerging adults and assist them in making decisions by offering advice on self-management and workplace accommodations. During this support process, an effort should be made to understand what goals the emerging adult attaches importance to. Also, collaboration among health care providers, employment counselors, and employers may facilitate successful

transition to employment for emerging adults with SB in Japan.

Struggles with transition to adult health care may be alleviated if the health care system had mechanisms to assist emerging adults with SB to plan and prepare for the transition. While there are transition support programs for individuals with SB in Japan (Date et al., 2008), the services are typically provided at large health care facilities, including university hospitals and pediatric care centers, and limited in number. To reach all individuals with SB and their families equally, the programs need to be available across Japan and easily accessible for individuals in both urban and rural areas. Nurses and other health care providers at the transition support programs can assist emerging adults with SB by helping them identify adult health providers and learn to assume adult role in health care setting (Spina Bifida Association, 2018). In addition, emerging adults' concerns about future health status and receiving adult health care should be discussed.

The fact that participants perceived lack of support from peers with the same condition is also an important point to consider. Lindsay et al. (2017) pointed out importance of social support from peers in transition to post-secondary education and employment. In addition to sharing information useful for successful transition to adulthood, emerging adults with SB may feel understood by communicating with peers in the same situation (King, Willoughby, Specht, & Brown, 2006). Health care providers' support with matching emerging adults with a peer mentor may be useful.

### **Recommendation for Future Research**

At present, no transition support programs for individuals with SB in Japan appear to be informed by theories or developed based on research findings. One of the priorities for future

research in transition for this population is to develop and test interventions to facilitate transition to adulthood and adult health care. The findings of this study will contribute to research process for developing and testing the interventions that will include identification of areas for interventions and development of an instrument to assess transition readiness for emerging adults with SB that takes Japanese social context into consideration.

In addition, there is a need to better understand the challenges of emerging adults with SB in Japan as well as their needs and resources. Since there is a wide variation in how individuals may be affected based upon the severity and physical limitations of the condition, research with emerging adults with milder form of SB will be needed.

### **Conclusions**

This qualitative descriptive study explored experiences of emerging adults with SB in Japan during their transition to adulthood. The study findings showed that the emerging adults with SB in Japan struggled with finding a job, establishing relationship with friends, implementing self-management, or making transition to adult health care. While the themes identified in this study were generally consistent with previous research, emerging adults with SB in Japan had unique experiences in the Japanese social context. Programs facilitating transition to adult life and adult health care based on research and guided by theories will be needed.

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### **Conflict of Interest**

The authors have no conflict of interest to declare.

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Table 4.1.

*The Demographic and Clinical Characteristics of Participants*

<b>Characteristics</b>	<b>n (%)</b>
Demographic characteristics	
Gender	
Male	5 (62.5)
Female	3 (37.5)
Age (years)	
20-24	4 (50)
25-29	4 (50)
Education	
High school	2 (25)
2-year college	1 (12.5)
4-year college	5 (62.5)
Employment	
Full-time	3 (37.5)
Part-time	3 (37.5)
Unemployed	2 (25)
Clinical characteristics	
Hydrocephalus	5 (62.5)
Self-management behaviors	
Self-catheterization	7 (87.5)
Bowel program	
Large volume enema	4 (50)
Other	2 (25)
Means of mobility	
Wheelchair user	6 (75)
Ambulates with orthosis	1 (12.5)
Ambulates without orthosis	1 (12.5)

Table 4.2.

*Themes, Sub-Themes, and Exemplar Quotes Describing Participants' Experiences during Transition to Adulthood*

Themes	Sub-Themes	Exemplar Quotes
Struggling	Struggling to find a job and work	<p>I guess I failed [employment interviews] because I wrote in the documents that I had bowel and bladder dysfunction and that I couldn't stand- --I couldn't stand at all and I leaked my urine and stool because of the level of my Spina Bifida lesion--- and so I wrote that I wanted the company to equip the workplace with a chair and that I also needed a bed, so I guess I failed because I told the people at regular companies about those things. So, I failed all the employment exams probably for that reason. (321-328, 03)</p> <p>Well, I worked there twice a week, and a coworker gave me a ride home, and there was no set end time for my job. I did it for about two years, but I got a pressure sore during the time I was doing it, and it was pretty much... I've been having pressure sores repeatedly since high school, and I got a very large pressure sore then, it was my fault because I was hiding it, though. So, I needed to rest at home for a month and a half. And, that was one reason that I quit the job. (330-336, 04)</p>
	Struggling with relationship with family	<p>But, regarding how much I learned to be thankful to my mother, there was a big difference between experiencing [doing things by myself at the dormitory] and not experiencing it. (124-126, 04)</p> <p>I can't leave my parents' house because I can't earn enough money, and as a result, I have to avoid causing trouble or I will put myself in a difficult position in the family later. (584-586, 08)</p>
Finding the Meaning of Being an Adult	Trying to achieve what the common definition of "being and adult" means	<p>Well, I define "an adult" in my mind as a person who is free and can take responsibility for anything he does, (421-422, 01)</p> <p>Well, becoming independent means, I think I'll have to think about my health, and well... I still live with my parents, but I think, when I live on my own I'll have to think about managing money, well, cooking in everyday life and stuff like that, I'll have to think about what I should do with those things, and therefore I think it's going to be tough. (205-209, 07)</p>

	Exploring the meaning of “being an adult” for a person with spina bifida	<p>When [the people from a company asked me during the job interview], “What does independence mean to you?” and something like “Why are you searched for a job?” I remember saying, “Because I want to become independent.” Some people said that [they were searching for a job] for their mothers or their families, but, well, I remember thinking something like, maybe [I was searching for a job] to become independent. However, I felt that it wasn’t actually true when I did get a job, hahaha (laughs), because I live with my mother. But, well, I’m in the environment where it’s possible to live on my own if I want to, but I’m getting negative about it, or I mean, I don't want to do so. (766-778, 03)</p> <p>I think that becoming an adult means becoming able to take care of yourself, like get around by yourself. And, I think that you are an adult if you learn about yourself and make decisions by yourself in the future. (332-334, 05)</p>
Learning to Implement Self-Management	Learning to implement self-management behaviors	<p>When I had a fever, it led to a complication, or it might lead to it, and I have to get dialysis in the worst scenario, so my parents were nagging me about it, and so now I go to bathroom properly. (464-467, 04)</p> <p>Well, it was the doctor I had been seeing that had the biggest impact. He said to me, "It's high time you learned to do catheterization by yourself!" (385-386, 08)</p> <p>The nurses have been telling me that it's better to learn to perform the bowel program by myself, but I don't have enough time to do it, and so I still rely on my mother for it. (277-279, 05)</p> <p>I can't do the enemas without my parent's help. I can't feel my buttocks, you know, so, even if I try to put the liquid [into the rectum], I don't have the slightest clue where [the catheter tip] is. It's impossible in the first place, I mean, I can't even insert the liquid, and so I rely on my parents for that at this point. And, I'm trying to find a way to [do enemas by myself], but I haven't found any concrete measures yet. (374-379, 08)</p> <p>Well, my health status is, I don't get sick or something, but there are times when I find my urine cloudy, and in those cases I get concerned that the catheterization might not be done hygienically. (L. 253-255, P05)</p> <p>In general, I go to the bathroom about once every three to four hours, but I went to the bathroom before I worked outside the office, and also, I tried my best not to drink water during the outside work. Well, I’m careful with urinary tract infections still now. Even now, I get it twice a year, I mean, I get urinary tract infections. Well, I get it twice a year, or three times a year when I get it more often. Well, I try to drink 1 to 1.5 liters of water per day. (429-440, 04)</p>

	Learning to seek help when necessary	Well, the school nurse was the one I turned to when I leaked accidentally (495-496, 03)
	Preparing environment for self-management	<p>I haven't got pressure sores for the past year, but I can get them anytime and anywhere because my body is like this, you know. So, I try to spend about 10 to 15 minutes lying on the bed at lunch break. I'd visited the office [before joining the company] and well, I joined the company after doing practical training there, and I had been asking them [for my use of the bed in the office] since the visit and they accommodated the bed, so I can work there. (357-362, 04)</p> <p>So, my mother tells the care workers how to help me with those tasks in case that I have to do the large volume enema when my mother is not at home. So, my mother arranges those things for me so that I don't get lost when she isn't at home. (619-622, 03)</p>
Health Care	Transferring to adult health care	Well, I used to go to [hospital], where I used to see the neurosurgeon as well, and I also used to see the ophthalmologist at the hospital, but I went to the ophthalmologist only once a year. I have strabismus, and so I only went there to monitor the progression of my strabismus ---I guess I used to get the checkup once a year in summer vacation. But, my strabismus is not progressing so rapidly and there is no problem with it, and therefore I started going to a clinic that is in my neighborhood and is closer to my place. It's an eye clinic that is closer to my house. It's a normal clinic and I go there when something comes up. (563-572, 03)
	Worrying health status and receiving health care	<p>I'm worried over my spinal deformity, I've been told that I should get surgery for it, but I was told that I won't be able to take care of myself anymore and I'll never be able to play sports if I get the surgery, so I've been hesitating. However, I'll definitely have to get the surgery if the curve gets worse, so it's scary to me. (396-399, 05)</p> <p>Well, the providers at the rehabilitation hospital where I receive catheters don't have expertise [in Spina Bifida], and therefore they can't understand when I talk about my condition in detail. They ask me the same questions every time, like "How many times do you do catheterization?" and stuff like that, but they can't make any concrete suggestions because they don't have the expertise. This condition doesn't get worse, but there is no solution for the issue "Who can I turn to in case it gets worse?"---I'm a bit worried about it now. (464-478, 08)</p> <p>For example, well, if I'm taken to a hospital in an ambulance, and if I hurt my leg and need to have surgery—I'll be troubled if I get anesthetic for lower body—I need to have general anesthesia done in such a case. ... Well, if I'm conscious, I can say "Take me to [hospital]," well, there is a hospital where</p>

		I get checkups, so I wish I can say “Take me to the hospital.” (321-335, 06)
Processes and Challenges in Establishing and Retaining Relationship with Friends	Nurturing relationships with friends at school/in community	<p>My friends said at the elementary school reunion, although I had disability like this, like they were glad to have met me, like I inspired them to try their best. (20-22, 07)</p> <p>I still have some childhood friends who don’t have disabilities and they have been with me since elementary school or kindergarten, and I still stay in touch with them, so I think it was a good thing. Also, in terms of networking, it’s nice to be able to meet people with many different personalities. (383-387, 01)</p> <p>Probably I’m spending the most fulfilled days with my friends from high school now, those friends from the high school [band] club let me have quite an experience. I said earlier in the interview, “Of course I go on a trip with my friends” and the friends [who go on a trip with me] are two of my friends from the high school [band] club. We went on a trip during summer vacation last year. (693-698, 03)</p> <p>Of course, I had friends I didn’t get along with, but it’s like I welcome people with open arms, but it was unavoidable after all that there were some people who focused on my body, my disability, and decided to leave me, but it’s like, I focus on how I can promote the friendship with the friends who are with me now. (726-731, 03)</p>
	Nurturing friendships with peers with spina bifida or physical disability	My peer in the [club] was living in the dorm, too, and so we talked a lot about the [club] and many other things, well, the friend and I were getting along well in our personal life. And sometimes, we talked about going somewhere and about doing something for fun, and stuff like that. (149-153, 04)
	Spending time with friends	<p>I really appreciate it when I enjoy myself with [my close friends from high school], and there was once a time when I said to them, “I’ve had a lot of great experiences,” and then they just seemed comfortable saying, “Why don’t you do what you think is fun?” So, I really like it. (737-740, 03)</p> <p>I chat with the friends [while I’m playing the sport] and take a trip with them, and stuff like that. (64-65, 05)</p>
	Struggling to be involved in romantic relationship	I hope I’ll be able to meet other people--- people outside my work, and like I’ll meet a nice person and get a boyfriend. Hahaha. (Laughs) (243-244, 03)

	Lack of peer support	I hope that I can connect with more people with the same condition and talk with each other. (410-414, 05)
Broadening Scope of Experiences	Creating opportunities by learning to get around on one's own	I would just go to school and go straight home when I was in elementary school and junior high because [my parents] would bring me to school and pick me up then, but after I entered high school, I was able to have my personal time to go shopping and something like that because I was able to go to school by myself. (109-113, 07)
	Barriers to broadening scope of experiences	There aren't places to hang out near the university, and so there are times when I give up chances to hang out with my friends because there is no accessible transportation when they plan to go somewhere far [from the university]. (225-227, 05)

## **CHAPTER FIVE**

This study focused on the experiences of transition for emerging adults with spina bifida (SB) in Japan. In this chapter, synthesis of all the manuscripts is provided. State of the science on the experiences of transition for this population is summarized from the two syntheses of literature: (a) synthesis of literature on transition experiences of adolescents and emerging adults (AEA) with SB written in English and (b) synthesis of literature on transition published in Japanese journals (and with one exception written in English). Reflections on research methods are also discussed. Next, a summary of the findings of the current study is provided. Finally, implications for theory, practice, research, and policy are discussed.

### **Synthesis of Manuscripts**

Advances in management of SB have led to remarkable improvement in the prognosis of children with the condition. However, individuals with SB confront various challenges as they grow into adulthood. Previous research has shown that acquisition of select self-management skills is delayed in adolescents with SB (Davis et al., 2006). Further, AEA with SB were found to experience psychosocial issues including difficulty with social adaptation, social isolation, and stigma (Kinavey, 2007; Ridosh et al., 2011). While it was assumed that emerging adults with SB in Japan had similar challenges and support needs during transition to adulthood, research studies conducted both in Japan and in other countries were reviewed to identify gaps in the literature concerning AEA with SB.

### **State of the Science from Two Syntheses of Literature**

Two syntheses of literature on transition to adulthood for AEA with SB were conducted to identify knowledge gaps in this area. The syntheses of literature both in Japan and in other

areas of the world, including North America and Europe, were conducted, because comparison of research findings in Japan with those in other countries helped to understand the state of the science and knowledge gaps on transition for AEA in Japan. The manuscript #A, titled “The transition Experiences of Adolescents and Emerging Adults with Spina Bifida: A Synthesis of Literature Written in English” provided synthesis of research literature written in English on transition to adulthood for AEA with SB. The topics relevant to the experiences of AEA with SB included emphasis on self-management behaviors (Ridosh et al., 2011; Sawin et al., 2009), independence (Ridosh et al., 2011; Sawin et al., 2009), psychosocial challenges including limited social interaction, discrimination, and stigma (Lindsay et al., 2017; Ridosh et al., 2011; Sawin et al., 2009), self-advocacy (Ridosh et al., 2011; Sawin et al., 2009), and exploration of identity as a person with disability (Kinavey, 2006, 2007). Studies revealed current conditions regarding transition outcomes including self-management (Bellin et al., 2011; Buran et al., 2004), functional independence (Buran et al., 2004; Heffelfinger et al., 2008), and social participation (Bellin et al., 2011; Dicianno et al., 2009; Liptak et al., 2010) as well as challenges to achieving those outcomes. Adolescents with SB were found to have challenges to engaging in some activities at a developmentally appropriate level including decision-making, responsibility, independence in self-management behaviors, and activities with peers (Buran et al., 2004). Also, more than 60% of young adults with SB lived with family (Woodward et al., 2012).

Further, research has addressed experience of transition to adult health care including barriers, facilitators, and solutions of health care transition in European and North American countries (Berry et al., 2013; Grimsby et al., 2016; van Staa et al., 2011; Young et al., 2009). There were also multiple research projects that developed and evaluated transition preparation

programs using experimental or quasi-experimental designs (Betz et al., 2010; Sawin et al., 2015).

The manuscript #B, titled “The Transition Experiences of Adolescents and Emerging Adults with Spina Bifida: A Synthesis of the Literature Published in Japanese Journals” provided the current state of the science on transition to adulthood for AEA with SB published in Japanese Journals. Previous research addressed challenges to performing self-management behaviors including medications (Hori et al., 2002), bowel programs (Domae et al., 2014; Hori et al., 2002; Kihara & Suzuki, 2008; Koike, 2007; Take, 2004), catheterization (Domae et al., 2014; Hori et al., 2002; Kihara & Suzuki, 2008), skin care (Hori et al., 2002; Kihara & Suzuki, 2008), and menstrual hygiene management (Noda et al., 2013). In a qualitative study on psychological and social process in which AEA with SB leaned to perform bowel management behaviors, Koike (2007) found that individuals with SB became increasingly aware of their difference from others as they acquired bowel management skills. Koike (2011) further revealed how women with SB attempted to obscure the difference from others and coped with their challenges, maintaining their self-esteem. Another theme explored in previous literature was issues related to social adaptation. Adolescents and emerging adults with SB in Japan had experiences of stigma and school bullying especially when they received education in regular school (Koike, 2008). In addition, research on mental health of AEA with SB in Japan revealed that the mean score of subjective well-being was significantly lower in young adults with SB than in those without the condition (Fujita, 2010). Further, the number of friends who have SB, feelings of belonging to the society, and feelings of trust were positively associated with subjective well-being in young adults with SB (Fujita, 2014).

Previous research also explored health care needs of AEA with SB in Japan. The majority of individuals with SB visited two or more specialists at a time due to complexity of their health needs (Date & Ito, 2003). In addition to health concerns related to the condition including bowel/bladder management, shunt status, and urinary tract infection, individuals with myelomeningocele, the most severe form of SB, had concerns and support needs related to education and employment (Date & Ito, 2003; Oi et al., 1996). As a solution for the patients' burden of receiving health care at multiple clinics and unmet needs, introduction of a multidisciplinary program that provides comprehensive care services including preparation for transition to adult health care was proposed (Date et al., 2008; Date et al., 2005).

In both of the syntheses of literature, it was found that AEA with SB confronted various social and psychological challenges during transition to adulthood. A qualitative descriptive study was needed to better understand the experiences and unmet needs of these AEA in the context of Japanese social and cultural context. Thus, the purpose of this study was set to describe the experiences of transition to adulthood for emerging adults with SB in Japan.

### **Reflections on the Methods**

There were several components of the methodology used in this study that add the potential to influence recruitment, data collection and perhaps the results. A screening tool, called "Cognitive Evaluation to Sign Consent Form" (See Appendix C) was used to assess the ability of the participants to understand the purpose and procedure of the study and provide consent. This assessment for comprehension was conducted because, some emerging adults with SB have intellectual disabilities or changes in learning or neuropsychological process (e.g., executive functioning, working memory, or processing speed) that might impair their ability to

consent (Dennis & Barnes, 2010; Fletcher & Brei, 2010). This tool was translated into Japanese from a previous study of young adults with spina bifida. This procedure may have placed some time burden on the participants; however, it helped to assure that the participants provided informed consent to participate in the study after fully understanding the researcher's explanations.

The researcher was unable to conduct member checking due to time constraints and participant availability issues although consent for a member checking session had been obtained from all the participants. As a result, there was no opportunity to confirm the themes from the perspectives of participants. This may have decreased the credibility of the research findings (Lincoln & Guba, 1985).

In addition, all the interviews were conducted in Japanese because, it was the first language for the participants. All interviews were transcribed initially into Japanese by the first author. Out of eight total interviews, four interviews were then translated into English by the first author before coding. The translation was confirmed by an expert in English translation of study data. This expert also had experience in qualitative research in sociology.

Each of these four translated interviews were read by two additional team members who confirmed the themes identified. The remaining four interviews were coded directly from Japanese transcripts. While data were analyzed in collaboration with two researchers in nursing who had extensive experience in qualitative research and in research on self-management and transition of individuals with SB, this data analysis may have negatively influenced the confirmability of the findings, since the team members only confirmed codes and themes written in English. Also, the first author was born and raised in the Japanese society; therefore, the

analysis by the first author was conducted from the perspective of a member of the community.

### **Qualitative Study on Experiences of Emerging Adults with Spina Bifida in Japan**

The third manuscript titled “Transition to adulthood: Experience of Youth with Spina Bifida in Japan” is the report of the findings of this qualitative descriptive study. Semi-structured interviews were conducted with eight emerging adults with SB 20 and 29 years of age living in Japan. Six core themes were identified from the interviews: (a) struggling, (b) finding the meaning of “being an adult”, (c) learning to implement self-management, (d) health care, (e) processes and challenges in establishing and retaining relationship with friends, and (f) broadening scope of experiences.

The findings highlighted “struggling” as an overarching theme. Consistent with previous studies (Koike, 2011; Lindsay et al., 2017), participants struggled with finding a job and working. They were faced with barriers, including employers’ reluctance to hire individuals with disabilities, lack of legitimate appraisal of their ability, inaccessible work environment, and internal culture of unwritten rules at the workplace that did not fit the participants. Further, some participants had challenges with protecting their own health while working due to lack of understanding or accommodation of their special health needs at workplace.

The perspectives participants shared also highlighted their exploration of the meaning of “being an adult” uniquely for a person with SB. Some participants described what they felt about their actual life situations when compared to the goals typically set during transition to adulthood. They did not necessarily try to achieve those typical goals. Rather, they explored what “being an adult with SB” meant for themselves based on their own experiences of living with the condition.

Participants shared their challenges related to transition to adult health care. They described their experiences of transferring to adult health care or to health care services different from the ones they used in their childhood. Some participants experienced sudden transfer to adult health care without sufficient planning and preparation. While transition preparation programs for individuals with SB have been developed in some pediatric hospitals in Japan (Date et al., 2008; Date et al., 2005), there appeared to be limited opportunity for support for health care transition for the affected emerging adults.

### **Implication for Theory**

This study was guided by three theoretical frameworks: (a) Theory of Emerging Adulthood, (b) Ecological Model of Secondary Conditions and Adaptation, and (c) Individual and Family Self-Management Theory. In the results manuscript (“Transition to Adulthood: Experience of Youth with Spina Bifida in Japan”), only the primary theory “Theory of Emerging Adulthood” was addressed due to the length restrictions of manuscript. In this chapter, relationships of this study will be discussed as it related to all three.

### **Theory of Emerging Adulthood**

The findings of the current study generally supported the features that are proposed to be distinctive to emerging adults, namely, exploring identity, instability, being self-focused, and feeling in-between (Arnett, 2015). However, unlike the emerging adults’ prospect for the future proposed in the theory (Arnett, 2015), the participants of the current study did not express their feelings that they possessed possibilities or were optimistic about their own future. Rather, they struggled to achieve developmental milestones for adulthood with uncertain future prospects. In addition, the participants appeared to explore what “being an adult” meant for themselves

while a person with SB and did not necessarily attempt to achieve typical goals for adulthood. This incongruence with the theory may be due to mixed contextual factors, i.e., challenges to achieving developmental milestones for individuals with the condition (Davis et al., 2006) and Japanese social and cultural context. A Japanese psychiatrist Takeo Doi (2001) analyzed the characteristics of Japanese people's interpersonal relationships using a unique Japanese word *amae*, which bridges the concepts of "dependence" and "attachment" (Doi, 1992). In this social and cultural context, dependence on family tends to be generally considered acceptable. Thus, the theory may need to be applied with caution to research with individuals who have specific backgrounds including chronic conditions or cultural background different than that of North America.

### **Ecological Model of Secondary Conditions and Adaptation**

This model proposes that two different types of risk factors, i.e., condition-specific and neuropsychological factors, and protective factors, i.e., adolescent resilience, family resourcefulness, and health care adequacy, are associated with adaptation outcomes of adolescents with SB (Sawin et al., 2003; Sawin & Thompson, 2009) and was thought relevant to young adults. In the current study, participants reported their experiences related to physical health including bowel and bladder continence, self-management, and secondary conditions (e.g., pressure sores and urinary tract infection), mental health, and participation in social life (e.g., employment and participation in activities with friends) as essential elements in their lives during transition to adulthood. These findings imply that the adaptation outcomes proposed in the model are important for emerging adults with SB in Japan.

Consistent with the protective factors proposed in the model, the findings of this study

imply that communication efficacy and support from family positively affect transition outcomes for the emerging adults. In addition, the findings suggested that establishing relationship with friends could facilitate positive feelings and social participation in emerging adults with SB in Japan. Thus, this factor may have a potential to be an additional protective factor for adaptation outcomes in this population.

### **Individual and Family Self-Management Theory**

In this theory, an individual and family incorporate behaviors for facilitating management of the condition into daily activities of the individual or family. Self-management is proposed to have three dimensions: context, process, and outcomes (Ryan & Sawin, 2009; UWM Self-Management Science Center, 2017; UWM Self-Management Science Center Working Group, 2011). The findings of the current study may support importance of some of the contextual factors proposed in the theory, such as complex health care needs, access to care, and setting and provider transition. In the current study, participants shared experiences of transfer to adult health care without planning and preparation. Also, some participants reported difficulty in doing catheterization at workplace due to lack of co-workers' understanding; these barriers to effective self-management may be considered as major social environment factors that affect self-management outcomes in the emerging adults with SB in Japanese social and health care context.

In addition, in the current study self-management was found to constitute part of both the process and outcome of transition into adulthood for participants. For example, participants described the process in which they learned to implement self-management behaviors, including building the skills to perform the behaviors, learning to seek help, and prepare environment

where they were able to implement self-management effectively. Also, participants reported the level of self-management they had achieved during the transition, such as performing catheterization by oneself and assessing his/her own health status. Thus, the finding suggested that participants perceived the two dimensions of self-management, i.e., a process and outcome, as proposed in the theory (Ryan & Sawin, 2009; UWM Self-Management Science Center, 2017; UWM Self-Management Science Center Working Group, 2011).

### **Implication for Research**

This study was conducted to bridge the knowledge gap in transition to adulthood for emerging adults with SB in Japan. One of the most important areas for future research will be to develop and test interventions to facilitate transition to adulthood and adult health care. Health care providers assisting individuals with SB have developed long-term follow-up programs for individuals with SB that includes transition support (Date et al., 2008; Date et al., 2005; Hoshino et al., 2019). These programs appear to provide multi-disciplinary support for transition to adult life and to adult health care (Date et al., 2008; Date et al., 2005; Hoshino et al., 2019). However, there is a lack of evidence on the effectiveness of those programs for individuals with SB. In the current study, challenges in emerging adults with SB during transition to adulthood were identified in various domains, including employment, relationships with family and friends, self-management, and health care transition. These findings imply what the needs of these emerging adults are and will contribute to the development of future program of research on transition preparation programs.

The findings of the current study will also contribute to the development of instruments to measure self-management behaviors and readiness for transition to adulthood for emerging

adults with SB in Japan. Several instruments for individuals with the condition during transition have been already developed and extensively used in North America, such as the Adolescent/Young Adult Self-Management and Independence Scale -AMIS II (AMIS II) (Sawin et al., 2018) and the Transition Readiness Assessment Questionnaire (TRAQ) (Wood et al., 2014). Adaptation of these instruments to the emerging adults transitioning in the Japanese social context or development of new instruments that capture variables relevant to the experience and needs of the emerging adults in Japan will be needed. The findings of this study may provide empirical basis for the development or adaptation of such instruments, which could be used to evaluate the effectiveness of transition preparation programs mentioned above.

In addition, empirical research on transition to adulthood for emerging adults with different types of SB will be needed to understand experience and needs of emerging adults with SB in Japan more fully. The findings of this study only reflect the perspectives and experiences of the participants, the majority of whom required a wheelchair for mobility. Therefore, research with emerging adults with milder physical limitations is needed. Future research should also be conducted with emerging adults with intellectual disability related to SB because there is a lack of research to explore the experiences of the population during transition.

### **Implication for Practice**

The findings of this study demonstrated the gaps in services to facilitate transition to adulthood and to adult health care for emerging adults with SB in Japan. The study suggested the emerging adults' struggle during transition, including barriers to employment and other components of adult life, as well as protective factors, such as relationship with friends and resourcefulness of family. Practice initiatives to alleviate the emerging adults' struggles and to

strengthen the protective factors during transition will be important to improve adaptation outcomes for the population.

Development of transition programs or initiatives to build skills, abilities and the performance of self-management behaviors is essential. In particular, half of the participants in this study reported their challenges with learning to perform bowel programs without family's help. Nurses and other health care providers assisting the emerging adults can help explore opportunities for building self-management skills and abilities earlier in childhood and adolescence, especially development and management of effective bowel regimen.

Consistent with previous studies (Koike, 2008; Ridosh et al., 2011), challenges with establishing and retaining relationship with friends during adolescence and emerging adulthood, including experiences of social stigma, were revealed. Many Japanese people's view on disability may have influence on participants' experience and the meaning attached to them. The religious traditions, i.e., Shinto and Buddhism, has affected the worldview of Japanese people (Stevens, 2013). As a result, disability was traditionally viewed as being related to negative karma (Stevens, 2013). Disability has been also viewed not only as an individual attribute, but also as a familial one (Stevens, 2013), thus placing both individuals with disability and their families at increased risk of social stigma. While there have been social movements for improvement in treatment of individuals with disability, this view may continue to have influence on how people perceive disability and an individual with disability in the Japanese society (Stevens, 2013). Health care providers need to pay attention to issues related to the adolescents and emerging adults' social relationships at school and work. Counseling, mental health assessment, and other professional services should be provided when appropriate to

prevent secondary conditions as suggested in the Guidelines for the Care of People with Spina Bifida and in Got Transition (Got Transition, 2021; Spina Bifida Association, 2018).

The study also has implications for practice to assist the emerging adults with SB in transition to adult health care. The participants' experiences of sudden transfer to adult health care implied lack of planning, preparation, and care coordination for transition for individuals with SB in Japan. Health care providers at the transition preparation programs can assist emerging adults with SB by helping them identify adult health providers, build self-management skills, and learn to assume adult role especially in communication with health care providers (Spina Bifida Association, 2018). Binks et al. (2007) conducted a literature review on transition to adult-centered health care for individuals with SB or cerebral palsy. They identified five key elements for positive transition: preparation, flexible timing, care coordination, transition clinic visits, and interested adult-centered health care providers (Binks et al., 2007). In addition, a systematic approach based on the use of evidenced-driven strategies for health care providers, individuals with a condition like SB and their families has been developed in the US (Got Transition, 2020). This collaborative has identified Six Core Elements of Health Care Transition 3.0, provides sample tools and has implementation guidelines (Got Transition, 2020). Both the five key elements identified by Binks et al. (2007) and the six core elements of Got Transition (2020) might be adapted by professionals in Japan who are developing transitions interventions or services that reflect the Japanese culture and health care system.

### **Implication for Policy**

This study has several implications for policy on employment and on transition to adult health care for individuals with SB in Japan. Most of the participants reported their experiences

of struggling to find a job and working. The governmental programs to facilitate employment for individuals with a disability and support their transition to employment include (a) legally mandated employment quota system to facilitate employment at regular companies (Matsui, 2013) and (b) employment transition programs either assisting individuals with a disability to build abilities and skills needed for employment or providing opportunities of work or other activities for people with disability (Ministry of Health, Labour and Welfare, n.d.). However, the findings of this study implied that in most cases both emerging adults with SB and their employers did not have sufficient information on these programs or access to professional support. Mechanism to guarantee access to the information and professional support for both the emerging adults and employers will be needed.

Another barrier to employment for emerging adults with SB in Japan is the lack of policy that supports paid caregivers to individuals with a disability thus facilitating their employment. Considering their limitations in mobility and complex health care needs, an amendment to the policy to enable utilization of caregivers at workplace at public expense will be beneficial for individuals with SB.

Policies on transition to adult health care may also need improvement. The Japan Pediatric Society (2014) proposed the principles for transition to adult health care for individuals who developed a chronic condition in childhood, emphasizing the importance of autonomy of the individuals and smooth transition based on the collaboration between pediatric and adult health care providers. Nevertheless, the current study showed that emerging adults with SB in Japan had limited access to support for transition to adult health care. To make transition preparation programs available at major health care facilities throughout Japan, creation of government

subsidies or modification of the medical service payment system to alleviate financial difficulty in offering the programs should be considered.

### **Conclusions**

This study explored the experiences of emerging adults with SB in Japan during transition to adulthood and to adult health care. The findings highlighted the emerging adults' struggles, including challenges with employment and relationships with family and friends, as well as possible protective factors during transition. Also, the emerging adults explored the unique meaning of "being an adult" for a person with SB in Japan. These findings were generally consistent with previous research. However, each finding reflected unique perspectives of the emerging adults transitioning to adulthood in the Japanese social and health care context. It is essential to assist the emerging adults to participate in social life and build self-management abilities and skills during transition to adulthood in a way that the individual's own perspective is understood and valued.

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APPENDIX A:

Demographic and Clinical Information Form (DCIF)

for Youth/Young Adults with Spina Bifida in Japan

I will ask you some questions about yourself, your family, and your condition and self-management. You may take breaks between the sections and when you become tired.

➤ **Questions about yourself**

1. Age \_\_\_\_\_

2. Sex F = 1 M = 2

3. Describe your living situation

1. Parent's home	5. Other _____
2. Your home	
3. Relative home	
4. Friend's home	

4. In school Yes \_\_\_\_ No \_\_\_\_  
If no, Year graduated \_\_\_\_ OR Drop out grade completed \_\_\_\_

- Indicate educational level:
- \_\_\_\_\_ Junior high school
  - \_\_\_\_\_ High school
  - \_\_\_\_\_ Technical school
  - \_\_\_\_\_ College Degree
  - \_\_\_\_\_ Advanced degree
  - \_\_\_\_\_ Other

Current grade in school if applicable \_\_\_\_\_

Indicate type of settings you receive(d) education:

- \_\_\_\_\_ Regular school
- \_\_\_\_\_ Special education
- \_\_\_\_\_ Both

5. Are you working? Yes \_\_\_\_ No \_\_\_\_

If yes,  
Type of work

- 1) Paid work
- 2) Volunteer work

Employment type

- 1) Full-time      How many hours per week do you work? \_\_\_\_\_  
2) Part-time      How many hours per week do you work? \_\_\_\_\_

\*You may take a break before going on to the next section.

➤ **Questions about your family**

6. Who is your family? Who is your family in addition to the members who you live with?

7. Your marital status

1) Single    2) Married    3) Divorced    4) Other
--

8. Do you have children?      Yes \_\_\_\_ No \_\_\_\_

If yes, number of children \_\_\_\_\_

9. Do you get extra help at home?      Yes \_\_\_\_ No \_\_\_\_

If yes, who is your main caregiver?

\*You may take a break before going on to the next section.

➤ **Questions about your condition and self-management**

10. Diagnosis

What is your diagnosis? \_\_\_\_\_

11. Do you have hydrocephalus?      Yes \_\_\_\_ No \_\_\_\_

12. Do you have shunt?      Yes \_\_\_\_ No \_\_\_\_

When was the shunt first put in \_\_\_\_\_ (in the first month of life or actual month of life)

Shunt revision    Yes \_\_\_\_\_      No \_\_\_\_\_

13. Number of surgeries \_\_\_\_\_

Specify types of surgery \_\_\_\_\_

14. Were you hospitalized in the last 12 months?      Yes \_\_\_\_ No \_\_\_\_

If yes, number of hospitalizations you have had in the last 12 months \_\_\_\_  
Reason(s) for hospitalization \_\_\_\_\_

15. Do you use a bladder program to manage your bladder? If so, tell me about the bladder program.
16. Do you use a bowel program to manage your bowel elimination? If so, tell me about the bowel program. What are you currently using and how does it work?
17. Do you use any program to manage your skin condition related to spina bifida? If so, tell me about the program.
18. Now I'd like to know about your mobility. Do you primarily walk, use a wc or some of both?

a) Mobility

\_\_\_\_\_ Walks without crutches

\_\_\_\_\_ Walks with crutches

\_\_\_\_\_ Walks with crutches but uses manual chair for sports or distances

\_\_\_\_\_ Walks with crutches but uses manual chair for primary mode of ambulation, may walk in home

\_\_\_\_\_ Manual wheel chair only

\_\_\_\_\_ Power wheel chair

b) Do you use Braces (i.e. AFOs). Yes \_\_\_\_ No \_\_\_\_

If yes, specify \_\_\_\_\_

19. How many times do you leave home a week?

Appendix B:

Interview Questions

- 
1. Could you tell me about your experiences during transition to adulthood?
- 
2. What are your feelings about growing into adulthood?  
What has it been like for you to grow into adulthood?  
If not addressed by the youth/young adult, ask:
    - What do you think about achieving independence?
    - Have you felt that there are various possibilities or that you have choices while you grow into adulthood? If so tell me about the possibilities or choices you see.
    - Could you tell me about your thoughts on and experiences of finding a job and working?
    - Could you tell me about your thoughts on and experiences related to building friendship and romantic relationship?
- 
3. Have you felt challenged in the process of growing into adulthood? If so, tell me about them?
- 
4. What are the positive aspects you have found in the process of growing into adulthood?
- 
5. What have been the main physical challenges of spina bifida for you? Have you had any health problems last year?
- 
6. Have you faced any learning challenges?
- 
7. Do/Did you need extra help when you are/were in school or at work? If so, who offers/offered help to you? What is/was helpful to you?
- 
8. What tasks have you needed to learn to care for your spina bifida? (e.g., Clean intermittent catheterization, bowel program, doing skin checks?)
- 
9. Is it important to you and your family for you to learn to do those tasks/behaviors on your own? If so, where are you in that process or how has it worked for you to learn to do these tasks yourself?
- 
10. Was there a change of health care settings between childhood and adulthood? If so, how did the change of care settings go?
- 
11. What does successful transition into adulthood look like for you?
-

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12. QOL questions:

We would like to know about how your thoughts about quality of life. Quality of life is a person's idea or perception of what his/her life is like, how his/her life is going. It can be affected by the person's physical health, psychological state, as well as culture, social relationship, and other factors. What things are important to your well-being?

First, I will read a few statements and ask you to give me a number between 0 (poor) and 100 (excellent) that BEST represents your current belief.

- 1) How would you rate your quality of life?
- 2) How would you rate YOUR parents' quality of life?
- 3) How would you rate YOUR FAMILY'S quality of life?

Next we want to know:

What makes up QOL for you?

For you what would make your QOL better?

For you what are the things that are important in being happy about life?

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13. Is there anything else you want to tell me about growing up into adulthood with spina bifida?

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APPENDIX D:  
University of Wisconsin-Milwaukee Institutional Review Board  
Notice of IRB Expedited Approval



**IRB#:** 18.311  
**Title:** Transition to adulthood: Experiences of Youth with Spina Bifida in Japan

**New Study - Notice of IRB Expedited Approval**

**Date:** December 27, 2018

**To:** Kathleen Sawin, PhD  
**Dept:** College of Nursing

**CC:** Shiho Murayama

**Leah Stoiber**  
*IRB Administrator*  
*Institutional Review Board*  
Engelmann 270  
P. O. Box 413  
Milwaukee, WI 53201 -0413  
(414) 229-7455 *phone*  
(414) 229-6729 *fax*

<http://www.irb.uwm.edu>  
[lstoiber@uwm.edu](mailto:lstoiber@uwm.edu)

After review of your research protocol by the University of Wisconsin – Milwaukee Institutional Review Board, your protocol has been approved as minimal risk Expedited under **Category 6 and 7** as governed by 45 CFR 46.110. Your protocol has also been granted approval to waive documentation of informed consent, as governed by 45 CFR 46.117 (c), for interviews conducted over the phone or Skype (or similar Internet-based product).

This protocol has been approved on **December 27, 2018** for one year. IRB approval will expire on **December 26, 2019**. If you plan to continue any research related activities (e.g., enrollment of subjects, study interventions, data analysis, etc.) past the date of IRB expiration, a continuation for IRB approval must be filed by the submission deadline. If the study is closed or completed before the IRB expiration date, please notify the IRB by completing and submitting the Continuing Review form found in IRBManager.


This study may be selected for a post approval review by the IRB. The review will include an in person meeting with members of the IRB to verify that study activities are consistent with the approved protocol and to review signed consent forms and other study related records.

Any proposed changes to the protocol must be reviewed by the IRB before implementation, unless the change is specifically necessary to eliminate apparent immediate hazards to the subjects. It is the principal investigator's responsibility to adhere to the policies and guidelines set forth by the UWM IRB, maintain proper documentation of study records and promptly report to the IRB any adverse events which require reporting. The principal investigator is also responsible for ensuring that all study staff receive appropriate training in the ethical guidelines of conducting human subjects research.

As Principal Investigator, it is also your responsibility to adhere to UWM and UW System Policies, and any applicable state and federal laws governing activities which are independent of IRB review/approval (e.g., FERPA, Radiation Safety, UWM Data Security, UW System policy on Prizes, Awards and Gifts state gambling laws, etc.). When conducting research at institutions outside of UWM, be sure to obtain permission and/or approval as required by their policies.

Contact the IRB office if you have any further questions. Thank you for your cooperation and best wishes for a successful project.

Respectfully,

  
Leah Stoiber  
IRB Administrator

APPENDIX E:  
Kobe University Institutional Review Board  
Notice of IRB Approval

様式18

平成 31 年 2 月 15 日

審査結果通知書

申請者  
看護学 領域  
講師 本田 順子 殿

神戸大学大学院保健学研究科長  
木戸 良明



【研究課題】 成人期への移行：日本における二分脊椎症のある若者の体験

【研究責任者】

所 属 看護学 領域  
職・氏名 講師 本田 順子

上記の実施計画について、審査の結果、承認と判定したので通知します。

記

【審査機関】 神戸大学大学院保健学研究科保健学倫理委員会

【承認年月日】 平成 31 年 2 月 15 日

【承認番号】 第 766 号

注)

1. 研究責任者は、毎年1回、「進捗状況報告書（別紙様式14）」を提出すること。
2. 研究責任者は、研究が終了した際には、「終了報告書（別紙様式15）」を提出すること。
3. 研究責任者は、臨床研究等に関連する重篤な有害事象及び不具合等の発生を知ったときは、速やかに必要な対応を行うとともに、直ちに「重篤な有害事象に関する報告書（第 報）（別紙様式16）」を提出すること。

\*The current study was approved by Kobe University IRB with Dr. Junko Honda as university PI and Shiho Murayama as co-investigator.

## CURRICULUM VITAE

Shiho Murayama

**Place of Birth:** Aichi, Japan

### **Academic Degrees:**

- 2021            University of Wisconsin-Milwaukee  
Doctor of Philosophy in Nursing  
Dissertation Title: Transition to adulthood: Experience of Youth with Spina  
Bifida in Japan
- 2002            University of Minnesota  
Master of Science  
Emphasis: Child/Family Nursing
- 1996            University of Tokyo, Japan  
Bachelor of Health Science

### **Professional Nursing Experience:**

- 2020-Present    Lecturer (part-time), Faculty of Nursing  
Kyoritsu Women's University, Tokyo, Japan
- 2015-Present    Lecturer (part-time), Faculty of Nursing and Medical Care  
Keio University, Kanagawa, Japan
- 2016-2018        Lecturer, Faculty of Medical Technology  
Teikyo University, Tokyo, Japan
- 2014-2016        Research Associate, Faculty of Human Sciences  
Sophia University, Tokyo, Japan
- 2006-2011        Research Associate, Faculty of Medicine  
University of Tokyo, Japan
- 2005-2006        Staff Nurse, Pediatric Unit  
Japanese Red Cross Medical Center, Tokyo, Japan
- 2003-2005        Research Associate, Faculty of Medicine  
University of Tokyo, Japan
- 2002-2003        Staff Nurse, Pediatric Unit  
Japanese Red Cross Medical Center, Tokyo, Japan

1998-1999      Staff Nurse-Midwife, OB/GYN Unit and Pediatric Unit  
Seirei Hospital, Nagoya, Japan

1997-1998      Staff Nurse-Midwife, OB/GYN Unit  
Daiyukai Daiichi Hospital, Aichi, Japan

### **Nursing Licenses**

1997-Present    Registered Nurse-Midwife, Japan

1996-Present    Registered Nurse, Public Health Nurse, Japan

### **Reviewer:**

2012-Present    Japan Journal of Nursing Science

2017-2019      Journal of Japanese Society of Pediatric Oncology Nursing

2013-2021      Journal of Japan Academy of Midwifery

### **Publications/Presentations**

Murayama, S., & Sawin, K. The family is invisible in a literature review of transition to adulthood for adolescents and emerging adults with spina bifida. Poster presentation. *The 14th International Family Nursing Conference, Washington, D.C., August 15, 2019.*

Murayama, S., Sawin, K. J., & Brei, T. J. Self-management perceptions of adolescent/young adults with spina bifida and their parents. Poster presentation. *The 10th International Family Nursing Conference, Kyoto, Japan, June 25, 2011.*

### **Professional Memberships:**

2010-Present    Japanese Society of Pediatric Oncology Nursing

2004-Present    Japanese Society of Child Health Nursing

2004-Present    Japan Academy of Midwifery

2004-Present    Japan Association for Research for Family Nursing

2003-Present    Japan Academy of Nursing Science

2001-Present    Sigma Theta Tau International